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At the Beginning and the End: End of Life Decision-Making for Extremely Preterm Infants

INAUGURAL-DISSERTATION
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Lay your sleeping head, my love,

Human on my faithless arm;

Time and fevers burn away

Individual beauty from

Thoughtful children, and the grave

Proves the child ephemeral:

But in my arms till break of day

Let the living creature lie,

Mortal, guilty, but to me

The entirely beautiful.

– W.H. Auden, “Lullaby”

Abstract

An extremely premature birth often comes unexpectedly to parents, which raises a series of difficult decisions regarding life-sustaining measures. The present thesis aims at providing more in-depth information on end-of-life decision-making for extreme preterm infants. In the general introduction I illustrate the background on neonatal intensive care for babies born extremely premature. This thesis is structured in three parts: a population survey, parental interviews and a theoretical reflection.

In the first part, the societal attitudes with regard to withholding or withdrawing care of extreme preterm infants in Switzerland are explored. A number of 1210 participants completed the survey (response rate 24%). Our findings revealed that the Swiss population highly valued shared decision-making. The socio-cultural background of participants influenced attitudes toward end-of-life decision-making. In addition, participants' self-rated solidarity was high towards extreme preterm infants who may develop disabilities. These findings contribute to discussions on standardizing highly specialized neonatal intensive care and on ethical aspects that underlie morally pluralistic societies. Moreover, the findings regarding solidarity have the potential to alleviate some pressure – regarding societal support and financial difficulties – on parents in the decision-making process.

In the second part, the experiences of parents of extreme preterm infants who were born and died at a University Hospital in Switzerland were explored. Participants included 20 parents of 13 extremely preterm infants (seven couples, five mothers, and one father) and five health care professionals. The main findings showed difficulties with parental bonding in the neonatal intensive care unit (NICU) and different recollections regarding parental involvement in decision-making. The results suggest that professional support can contribute to parents' perception of being a family, positively affecting the grieving process. It further illustrates the necessity of careful professional evaluation of parents' wishes about involvement and clear descriptions of medical facts and treatment options.

The third part, a theoretical reflection on policies that limit intensive care measures for babies at a certain gestational age is presented. The reasons for such policies are explored and the ethical justifications are questioned. It argues that such policies are unjust to premature babies and ought to be abolished. In their place, individualized treatment decisions for premature babies are proposed.

Finally, this present thesis leads to some potential ideas for future practice and research in neonatal ethics. Six suggestions are presented: (1) share decisions to continue or discontinue life-sustaining interventions; (2) further institutionalize palliative and bereavement care in the NICU; (3) investigate how to manage divergent attitudes and values; (4) explore whether transparent counselling is warranted in withholding treatment; (5) investigate practices at the limit of viability and; (6) question thresholds in policies for decisions on resuscitation.

In conclusion, although the beginning and the end of extremely premature babies are so close, we should support these infants and their families to make the decision that is right for the infant and his or her family.

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Table of Contents

Abbreviations

EoL	End of Life
EoLDM	End of Life Decision-Making
ELGANs	Extremely Low Gestational Age Newborns
EPBs	Extremely Premature Babies
EPIBEL Studies	Outcome Studies for Extremely Preterm Infants in Belgium
EPICure Studies	Outcome Studies for Extremely Preterm Infants in UK
EPIPAGE Studies	Outcome Studies for Extremely Preterm Infants in France
EXPRESS Studies	Outcome Studies for Extremely Preterm Infants in Sweden
GA	Gestational Age
HCPs	Health Care Professionals
HRQoL	Health Related Quality of Life
IC	Intensive Care
LSIs	Life Sustaining Interventions
NICHD	National Institute of Child Health and Human Development
NICU	Neonatal Intensive Care Unit
NDI	NeuroDevelopmental Impairment
NRN	Neonatal Research Network
PC	Palliative Care
QoL	Quality of Life

Extended Summary

An extremely premature birth often comes unexpectedly to parents, which raises a series of difficult decisions regarding life-sustaining measures. In fact, the care for each and every baby raises a set of different medical questions and involves different ethical concepts. The present thesis aims at providing more in-depth information on end-of-life decision-making for extreme preterm infants. In the general introduction I will explore neonatal intensive care for babies born extremely premature. I summarize the recent epidemiological findings on mortality and morbidity and outline and discuss the ethical concepts relevant to decision-making for extreme preterm infants. The present thesis is structured in three parts: a population survey, parental interviews and a theoretical reflection.

In the first part, the societal attitudes with regard to withholding or withdrawing care of extreme preterm infants in Switzerland are explored. This nationwide trilingual (French, German and Italian) telephone survey is one of the first and largest representative studies to analyse public perceptions and values toward extreme prematurity. It is also the first investigation of the notion of solidarity in relation to societal expenditures and intensive care decisions. A number of 1210 participants completed the survey (response rate 24%). Our findings revealed that the Swiss population highly valued shared decision-making. The socio-cultural background of participants – and especially their linguistic region – influenced attitudes toward end-of-life decision-making. In addition, participants' self-rated solidarity was high towards extreme preterm infants who may develop disabilities. These findings have two possible implications. First, these findings on socio-cultural divergences contribute to discussions on standardizing highly specialized neonatal intensive care and on ethical aspects that underlie morally pluralistic societies. Second, the findings regarding solidarity have the potential to alleviate some pressure – regarding societal support and financial difficulties – on parents in the decision-making process.

In the second part, we analysed the experiences of parents whose infants were born and died within a few hours or days after birth at a University Hospital in Switzerland. The study focused on how parents experienced the dying trajectory of their extremely premature infants in the neonatal intensive care unit (NICU), how parents experienced parenthood, and how end-of-life decisions took place. Participants included 20 parents of 13 extremely preterm infants (seven couples, five mothers, and one father). Parental perspectives were further complimented with qualitative interviews with five caregivers who reflected on their experience with counselling

parents. The main findings showed that parents experience a multitude of stressors due to the immediate separation after birth, the alienating setting of the NICU, the physical distance to the child, medical uncertainties, and upcoming decisions. Even though they are considered to be parents (assigned parenthood), they cannot act as primary caregivers. Instead, they depend on professional instructions for access and care. Embodied parenthood can be experienced only at the end-of-life, that is, during the dying trajectory and after the child's death. Furthermore, parents described factors that affected the decision-making process in satisfactory or unsatisfactory ways. Transparent information, empathy, and honesty enhanced communication between parents and the health care team. Lack of transparent information and continuous support decreased satisfaction. Parents recalled different levels of involvement in decisions. Few parents recalled being involved in the decision-making process, some parents experienced a dissociative state of mind that hindered their involvement, whereas others felt actively involved. Thus, our results first suggest that professional support during difficult process contributes fundamentally to parents' perception of being a family, which could support the grieving process. Second, this illustrates the necessity of careful professional evaluation of parents' wishes about involvement and clear descriptions of medical facts and treatment options.

In the third part, a theoretical reflection on policies that limit intensive care measures for babies at a certain gestational age is presented. The reasons for such policies are explored and the ethical justifications are questioned. Many experts now recommend that clinical decisions about the treatment of such babies be individualized and consider many different factors. Nevertheless, many policies and practices throughout Europe and North America still appear to base decisions on gestational age alone or on gestational age as the primary factor that determines whether doctors recommend or even offer life-sustaining neonatal intensive care treatment. These policies are well intentioned. They aim to guide doctors and parents to make decisions that are best for the baby. That is an ethically appropriate goal. But in relying so heavily on gestational age, such policies may actually do the babies a disservice by denying some babies treatment that might be beneficial. In this paper, we argue that such policies are unjust to premature babies and ought to be abolished. In their place, we propose individualized treatment decisions for premature babies. This would treat premature babies as we treat all other patients, with clinical decisions based on an individualized estimation of likelihood that treatment would be beneficial.

As for the implications, this present thesis leads to some potential ideas for future practice and research in neonatal ethics. There is still much to be learned in end-of-life decision-making for

extreme preterm infants about how parents and health care professionals make decisions in practice and on how these decisions should be conceived from an ethical perspective. First, parents should be given the opportunity to share in decisions to continue or discontinue life-sustaining interventions. Hence, parents should be continuously involved in re-evaluating the appropriateness of LSIs and whether parents want to share in the decision-making or not. It should be respected when parents prefer not to participate in the decision-making and wish to leave the final decision to the health care professional. Furthermore, it may be possible that some parents need more guidance in the beginning. Either way, HCPs should continuously inform parents and aim to ensure they understand the available medical information.

Second, the present thesis illustrates the importance of further institutionalizing palliative care (PC) and bereavement care in neonatology. Establishing a good quality of PC is of particular importance for parents of extremely premature infants since parental bonding has shown to be particularly difficult. The parents may only have a very short, but very precious time with their child since extremely premature infants may deteriorate and die more quickly than other patients. Bereavement and perinatal PC further strives to minimize parental feelings of regret on how they spent the time with their child whilst alive and dying.

Third, our findings have illustrated the need to investigate how to manage divergent attitudes and values of parents, health care professionals and society regarding extreme prematurity. When weighing the burdens and benefits of withholding or withdrawing life-sustaining measures, it is not always possible to avoid subjective judgments about harm, risk, and long-term benefit. Hence, improving the communication skills of HCPs might solve misunderstandings of the relevant facts. Although it is important to avoid potential misunderstandings, reasonable people might disagree on the basis of different values. Tools such as decision-aids or a communicative ethics may help to reduce conflict and establish a better understanding of different values between HCPs and parents. Hence, research should further investigate and improve these tools in order to include a recognition and deliberation of different experiences, attitudes and values between parents and HCPs.

Fourth, future research should explore whether transparent counselling is warranted in withholding treatment. Particularly, it is necessary to investigate how much information or disclosure should and can be given to parents. Although it is possible that nondisclosure happens with the best intentions, i.e. keeping the well-being of parents in mind, unforeseen disclosure may cause harm and must be taken seriously. Thus, it should be further explored how much information

should be shared within a shared decision-making process and a further exploration of the duty to disclose regarding gestational age policies is warranted.

Fifth, future observational or ethnographic studies are necessary to closely examine the practices and the management decisions at the limit of viability. Although concerns regarding gestational age policies are voiced by parents and clinicians and investigated in many questionnaire studies, little ethnographic data exists from clinical practice.

Finally, a normative implication of the present thesis is that thresholds policies for decisions on resuscitation should be questioned. It is necessary to continue to address and discuss the limits of gestational age estimations and local outcome data within the neonatal community to foster a change in the way physicians treat babies born extremely premature. It is furthermore important to realize that thresholds of care can change with time, technology, politics (i.e. national recommendations), economics and morality. Therefore, obstetricians, neonatologists, midwifes, neonatal nurses and ethicists should continuously reflect and debate on the boundaries of the grey zone, realizing that these boundaries are not static.

In conclusion, although the beginning and the end of extremely premature babies are so close, we should support these infants and their families to make the decision that is right for the infant and his or her family.

INTRODUCTION

Outline

The present thesis addresses questions around extreme prematurity in Switzerland, and is structured in three parts. Prior to illustrating these three study components, I will provide a background on neonatal intensive care for babies born extremely premature. A clear definition on extreme prematurity is presented and recent epidemiological findings on mortality and morbidity are summarized. Moreover, the ethical concepts relevant to decision-making for extreme preterm infants are outlined and discussed. Then, I present an overview of the original research conducted during this PhD project and an outline of the aims of all three parts of the study: a population survey, parental interviews and a theoretical reflection.

Extreme Prematurity – Medical Progress and Moral Controversy

A pregnancy usually lasts about 40 weeks from the first day of the woman's last menstrual cycle. A birth that takes place at 37 completed weeks gestation is a premature birth. Babies born before the 7th month of pregnancy, i.e. before 28 completed weeks of gestation are referred to as *extremely* premature. Common causes of a premature birth can include 'multiple pregnancies, infections, and chronic conditions, however, the cause is often not known' (WHO 2016). Although not always identical in meaning (see Table 1), it is important to note there are many different ways to refer to this patient population such as: extremely premature infants or babies (i.e. before 28 completed weeks); extremely low birth weight infants (i.e. ≤1000 grams); extremely low for gestational age newborns (ELGANs); babies born at the limit of viability (i.e. before 26 completed weeks); or preemies. For our studies, we defined extremely premature infants as those born before 28 completed weeks of gestation.[1]

[1] With the exception of the third study where I focus on infants born before 25 weeks of gestation. In this chapter, I analyze gestational age policies that affect care for babies born at these ages.

Table 1. Various Descriptive Terms for Delivery and Birth of Extreme Preterm Infants

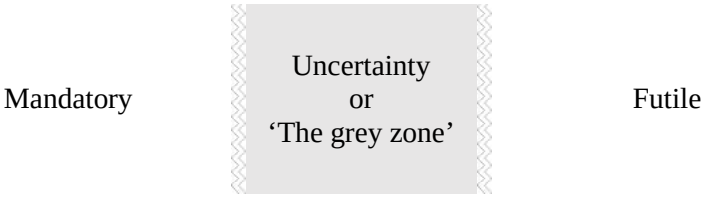
Birth Weight Specific Descriptions of Extreme Preterm Birth	
Very low birth weight	<1500 g
Extremely low birth weight	<1000 g
Micropreemie	Variably defined <1 ¾ lb or <3 lb, or <1000 g, or <26 or <29 wk of gestation
Gestational Age Specific Descriptions of Extreme Preterm Birth	
Midtrimester or second trimester	13th to 27th wk of pregnancy, 4th through 6th month of pregnancy
Remote from term	No consistent definition
Early preterm	No consistent definition
Extremely low gestational age	No consistent definition
Extreme preterm	<28 wk of gestation
Very preterm	28 through 31 6/7 wk of gestation
Newborn Outcome Specific Descriptions of Extreme Preterm Birth	
Previable	Not sufficiently developed to survive outside the uterus
Marginal viability	22 0/7 through 26 6/7 wk of gestation
Threshold of viability	At or before 25 wk of gestation or <750 g, 22–25 wk of gestation
Periviable	20 0/7 through 25 6/7 wk of gestation

* Adapted from Mercer (2017)

Extremely premature babies are often classified into three categories (see Figure 1). The first consists of patients for whom life-sustaining interventions (LSIs) are considered to be beneficial. Such infants have a reasonable chance for a good outcome. In the second category, interventions are determined to be futile. The infant is considered too immature to have a reasonable chance to survival without severe impairments. The difficult cases, then, fall into the third category, where infants are in-between the first two categories, i.e. where it is rather uncertain whether treatment is futile or beneficial. This 'in-between stage' is called the either the grey zone (for prenatal decisions) or the zone of uncertainty (for neonatal decisions) within which prognosis is uncertain.

Conventionally, patients in the first category receive LSIs, in the second they are offered comfort care and in the third group, decisions whether to provide intensive care are made on a case-by-case basis (Seri and Evans 2008, Dupont-Thibodeau, Barrington et al. 2014).

Figure 1. Three different categories that affect end-of-life decisions



In the present thesis I will mostly focus on cases in the 'in-between stage' and near the boundaries of the first and second category. The boundaries of these three categories vary depending on where the baby is born. This is often based on the available outcome data (usually national and local data) and is defined by the national societies of neonatology and obstetrics. The same holds true for the limit of viability. A foetus is viable when it is mature enough to survive into the neonatal period with the clinical support that is available (Seri and Evans 2008). Viability is a function of both biology (i.e. age, weight, race and gender) and biomedical technologies (e.g. antenatal corticosteroids, antenatal transfer to a level III NICU centre) and, as a consequence, is different in different parts of the world (Breborowicz 2001). These (inter)national variances will be highlighted more in-depth throughout the present thesis, but I will focus on the differences within Europe and North-America. More importantly, at present, there is no uniform gestational age that defines viability. Some scholars suggest the limit is at 21 or 22 weeks (Guillen, Weiss et al. 2015, Saugstad 2016).

A premature birth often comes unexpectedly. Only 0.3-0.4% of all babies are born extremely premature in Switzerland (Bundesamt für Statistik 2017). Over the past several decades, technological improvements in the field of neonatology have led to an increase in survival numbers for infants born at the margin of human viability. Overall, more infants survive today than 40 years ago (Verhagen and Janvier 2016). However, increased survival rates for extremely premature infants are not an unmitigated success. Some survivors are left with lifelong medical problems such as chronic lung disease, cognitive or developmental delays and neurologic impairments (Jarjour 2015). Prematurity is a distinct medical field; it is always an acute crisis with possible

complications and for some infants also a chronic condition. It is the acuteness of the condition that requires urgent medical interventions whereas the possible long-term complications necessitate a reflection on what constitutes best interest for this infant (Lantos and Meadow 2006).

Although these babies represent the cutting edge of neonatal medicine, many health care professionals, ethicists, policy-makers and parents debate about the (un)appropriateness of offering these children active intensive care (Pignotti and Donzelli 2015). Some experts have criticized neonatology's medical success as an example of medicine's moral ambiguity or hubris (Silverman 1992, Silverman 2004). The same goes for parents; some parents consider the medical team as the saviour of their infant and the NICU as a place where miracles can happen. Yet others see it as a place where parents are denied their rights to make medical and moral decisions for their child. As stated by Lantos and Meadow (2006, p. 5) 'these opposing interpretations of the NICU – modern miracle or medical nemesis – grip our collective moral imagination'.

The care for each and every baby raises a set of different questions and involves different ethical concepts. Are we doing more harm than is right (i.e. best interest)? Is death a greater harm than a lengthy stay in the NICU and a survival burdened with severe impairments (i.e. quality of life)? Once intensive care initiated, when is it enough, when can we withdraw LSIs (i.e. futility)? And, finally who should decide upon these questions (i.e. parental autonomy, physician authority and/or shared decision-making) (Lantos and Meadow 2006, Carter 2017)? Before delving into these questions and ethical concepts, it is important to present some facts relating to extreme premature birth and neonatal intensive care.

Epidemiology in the Neonatal Intensive Care

First of all, when referring to extremely premature babies born <28 weeks of gestation, it is important to note that obstetricians and neonatologists consider gestation (GA) by completed weeks and days. Accordingly, when a baby is born at 27 weeks of GA, the child is born somewhere between 27 weeks and 0 days (27 0/7) and 27 weeks and 6 days (27 6/7). Of note, gestational age is determined prenatally through the mothers last menstrual period and/or an ultrasound examination of the foetus during the first trimester. However, and with few exceptions, the precise GA cannot be known with certainty. The precision of gestational age determination by early ultrasound is ± 4 days (Sabbagha 1987, Berger, Bernet et al. 2011). If instead the history of the last menstrual period is used for GA determination, a much broader range of -6 to +14 days must be

taken into account (Wingate, Alexander et al. 2007, Berger, Bernet et al. 2011, Butt, Lim et al. 2014).

In the past decades, GA was used as the major predictor for mortality and morbidity. However, other factors also significantly influence outcomes in extremely preterm infants, such birth weight, sex, exposure or non-exposure to antenatal corticosteroids, multiple or singleton birth, type of delivery, and birth in a tertiary centre. Accordingly, the estimated probability of survival can vary considerably among infants around the same gestational age. For example, a female infant, 24 weeks GA, 750-grams, who received antenatal corticosteroids could have a better survival outcome than a male, 25 weeks, 525-grams, small-for-gestational-age who did not receive antenatal corticosteroids (Tyson, Parikh et al. 2008). Today, it is generally accepted among neonatologists to recognise and include all relevant prognostic factors into the decision-making process (Guillen, Weiss et al. 2015).

Over the last decades, several studies have aimed at describing the mortality, the morbidity and the chances of survival with or without impairment of extremely premature infants. Large epidemiologic studies have been performed worldwide, such as the EPICure studies (Extremely Preterm Infants Cure) in the United Kingdom, the EPIPAGE ('Etude Epidémiologique sur les Petits Ages Gestationnels') studies from France, the EXPRESS (Extremely Preterm Infants in Sweden Study) from Sweden, and many other population studies from Norway, the United States and Switzerland. Consequently, data from national cohorts or population based studies are used to counsel parents during decision-making in the perinatal and neonatal period (Marlow 2015). Let us know explore these mortality and morbidity outcomes.

Neonatal Outcomes of Survival

The survival rates of extremely preterm infants depict wide variation between countries (see Table 2 and Figure 2 for an overview of survival rates). The EPICure-2 cohort study from England in 2006 reported low survival rates for babies born <24 weeks of GA. Active intensive care was withheld for a large group of infants born at 22 weeks (73%), versus 16% at 23 weeks, and <2% for 24-26 weeks GA (Costeloe, Hennessy et al. 2012). Similarly, in the French cohort study EPIPAGE-2 no infants born at 22 weeks and only one at 23 weeks survived to discharge. Respectively, 90% of infants born at 22 or 23 weeks died after withholding or withdrawing intensive care in the delivery room (Perlbarg, Ancel et al. 2016). Survival rates in Switzerland are quite analogous to those of France, where mortality was 95% with most deaths occurring in the

delivery room for infants born below 24 weeks GA (Chen, Bajwa et al. 2016). Similarly, a recent study on causes and circumstances of deaths in extreme prematurity in Switzerland showed that most deaths of live-born infants in the delivery room died following primary non-intervention with relation to GA in a similar. In contrast, NICU deaths occurred following unrestricted life support regardless of GA (Berger, Steurer et al. 2017).

Table 2. Survival Rates (in %) to Hospital Discharge among Infants Born 22 to 25 Weeks' Gestation (% From Low to High).

Cohort	Year	Study	Denominator[a]	22 Wk	23 Wk	24 Wk	25 Wk
France, Epipage-2[1]	2011	Population based	Live-born infants	0	1	31	60
Switzerland[2]	2000-2004	Population based	Live-born infants	0	5	30	50
United Kingdom, Epicure-2[3]	2006	National cohort	Live-born infants	2	19	40	66
			Admitted infants	16	30	47	69
United States[4]	2006-2011	Population based	Live-born infants	5.1	23.6	54.9	72
			Admitted infants	23.1	33.3	56.6	72.3
Norway[5]	2013-2014	Population based	All births	5	17	41	65
			Live-born infants	18	29	56	84
			Admitted infants	60	35	58	86

Cohort	Year	Study	Denominator[a]	22 Wk	23 Wk	24 Wk	25 Wk
Sweden, EXPRESS[6]	2004-2007	Population based	All births	3.5	29	50	67
			Live-born infants	9.8	53	67	82
			Admitted infants	26	65	73	84

[1] Perlbarg, Ancel et al. (2016), [2] Fischer, Steurer et al. (2009), [3] (Costeloe, Hennessy et al. 2012), [4] Rysavy, Li et al. (2015), [5] Stensvold, Klingenberg et al. (2017), [6] Fellman, Hellstrom-Westas et al. (2009)

[a] All births include stillborn infants and refers to the survival rate of delivery; live-born infants refers to the survival rate of resuscitation; and admitted infants refers to the survival rate of babies admitted to the NICU.

Moreover, survival rates in the United States are incrementally higher for infants born at 22 or 23 weeks of gestation (Rysavy, Li et al. 2015). A similar trend is shown in other countries such as Sweden and Norway. Studies from Norway (2013-2014) and the Sweden (2004-2007) have reported higher survival rates for all births, all live-born infants and infants admitted to the NICU at <25 weeks (Fellman, Hellstrom-Westas et al. 2009, Stensvold, Klingenberg et al. 2017). In Norway, the survival rate among live born infants was higher at 18% at 22 weeks, 29% at 23 weeks, 56% at 24 weeks and 84% at 25 weeks (Stensvold, Klingenberg et al. 2017). In comparison[2], as shown in Table 2, the number of infants admitted to the intensive care is similar in Norway and Sweden, but more babies are likely to survive at 23 and 24 weeks in Sweden (Fellman, Hellstrom-Westas et al. 2009, Stensvold, Klingenberg et al. 2017). In the UK survival is almost non-existing for 22 weeks GA and is much lower at 23 weeks (Costeloe, Hennessy et al. 2012), whereas in France and Switzerland infants almost never survive before 24 weeks, and even at 24 and 25 week their survival rates were lower (Fischer, Steurer et al. 2009, Perlbarg, Ancel et al. 2016).

[2] Important to note is that, these comparisons are complicated by the fact that these data are reported by various countries with different sample sizes, populations, and time periods. These points are further discussed in paragraph 'Divergences in Neonatal Outcome Statistics'.

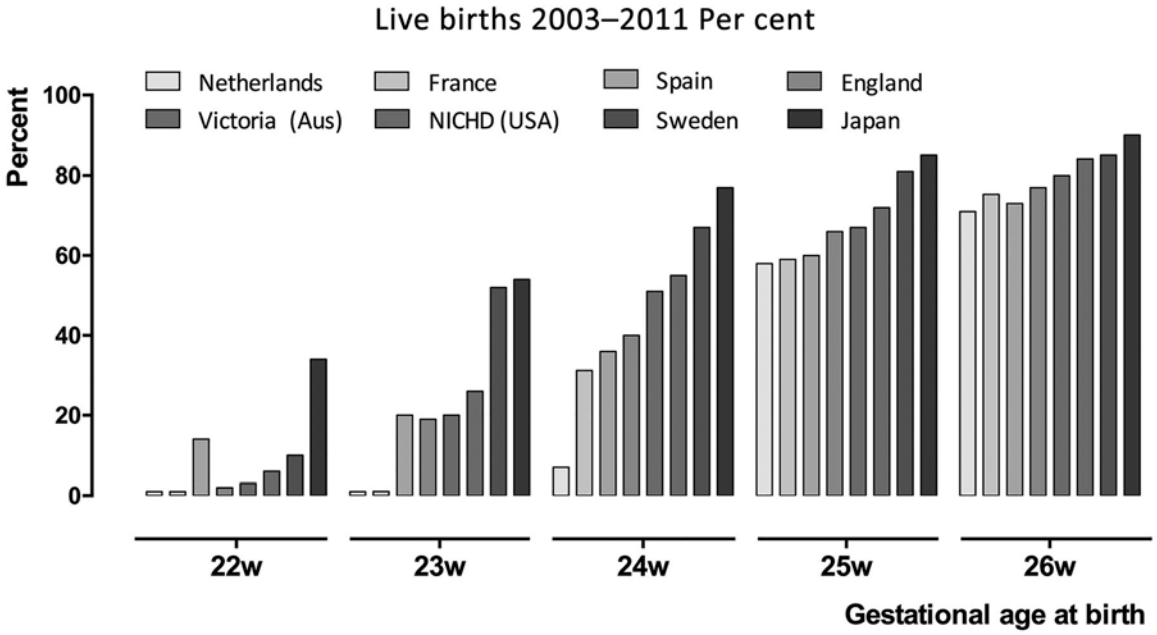


Figure 2. Survival of live births in large population-based studies 2003–2011. NICHD, National Institute of Child Health and Human Development; w, weeks. Figure adopted from Johnson and Marlow (2016).

Besides these wide variations between countries, there is also much variation in practice between regions and individual hospitals within a given country. Take for example the results from several population studies in North America and Europe. The Eunice Kennedy Shriver National Institute of Child Health and Human Development (NICHD) Neonatal Research Network (NRN) in the United States has analysed data (2006-2011) collected in 24 hospitals – of various sizes and in several U.S. regions, with diverse patient demographics, clinical practices, and outcomes – to identify variation in hospital rates of active treatment. This study has shown that the selection of active treatment differed for infants born <25 weeks GA. For babies born at 22 weeks, active intervention policies in hospitals ranged from 7.7 to 100%. This means that some hospitals treated all 22-weekers, and some did not provide care for infants born at 22 weeks GA. The percentage increased for babies born at 23 weeks with an active intervention rate that ranged from 52.5 to 96.5% between hospitals. Finally, almost all infants born at 24 weeks received intensive care with a range of 95.2 to 100% (Rysavy, Li et al. 2015). Regional differences have also been reported in Sweden, where mortality rates were predominantly influenced by local policy practices for delivery and management immediately after birth (Serenius, Blennow et al. 2015). Additionally, a Swiss study has showed that the medical centre where care was provided was a highly significant predictor of outcome (Steurer, Adams et al. 2015). A more recent Swiss study was unable to

confirm or contradict these results due to the study's limited sample size that did not allow for centre-to-centre comparison (Berger, Steurer et al. 2017). Let us know turn to the long-term developmental outcomes of infants born at the limit of viability.

Long-Term Developmental Outcomes

Despite the improved survival rates for extremely premature births, the chances of an extremely premature infant to survive with severe long-term neurodevelopmental impairment (NDI) plays an important role in the decision-making. A possible severe NDI can impact the decision whether to withhold treatment at the time of birth or to proceed with intensive care once admitted to the NICU (Moore, Lemyre et al. 2013). Currently, data on NDI is not homogenous and often varies widely between studies. Some studies report decreasing numbers of neurodevelopmental impairments (Doyle, Roberts et al. 2010, Moore, Hennessy et al. 2012), while others observe unchanged (Tommiska, Heinonen et al. 2007), or increased neurodevelopmental disabilities (Wilson-Costello, Friedman et al. 2005, Claas, Bruinse et al. 2011).

A recent meta-analysis of neurodevelopmental outcomes in cohort studies gives an insight on the relation between gestational age and NDI. The study shows there was a decrease in moderate to severe NDI for each increasing GA week. However regardless of GA at birth, a high percentage of survivors have a likelihood of developing moderate to severe NDI (Moore, Lemyre et al. 2013). Other studies have also shown little difference between each GA week and severe long-term impairments (Andrews, Lagatta et al. 2012, Meadow, Lagatta et al. 2012). For example, infants born <24 weeks GA did not have a worse moderate or severe long-term outcome than children born at 24 weeks GA. This supports the claim that gestational age is not a good predictor for long-term NDI of extremely premature babies born between 22-24 weeks (Herber-Jonat, Streiftau et al. 2014). Such evidence could affect decision-making for infants born at the limits of viability. As stated by Anderson et. al. 'if mortality in the NICU is the outcome that parents most fear, then physician counselling and public policy pronouncements that rely strongly on gestational age are epidemiologically and ethically appropriate. However, if survival of an infant with severe neurologic impairment is the outcome most feared, then reliance on gestational age appears to be misplaced' (Andrews, Lagatta et al. 2012).

These two paragraphs illustrated the heterogeneity of mortality and morbidity rates of live born infants <25 weeks' gestation. There are several explanations for these divergences in neonatal survival and long-term outcomes, which will be outlined in the next paragraph.

Divergences in Neonatal Outcome Statistics

In this paragraph, I will raise two explanations as to the divergences in neonatal outcome statistics. First, the different factors that result in divergences in rates of delivery, live birth admission to the NICU and survival will be highlighted. These variations can result into 'major differences in reported survival and, consequentially, have large effects on apparent rates of adverse long-term outcome' (Smith, Draper et al. 2014). Second, outcome statistics are also influenced by the attitudes and values of the different stakeholders.

Defining the Cohort

Reported survival rates are affected by several key issues, such as selection of various dominators, study type, thresholds regarding active life-sustaining treatment, and individual outcomes. On a side note, it is also important to recognize the difficulty and potential bias when comparing different studies and, thus different sets of outcome data, when these studies are conducted in different years and/or decades (Stensvold, Klingenberg et al. 2017).

First, the use of different denominators for calculating survival leads to bias in reported survival and long-term outcomes. There are four common denominators used in these studies such as all births (including stillbirths), live births, infants admitted to the NICU, and infants discharged home. Each denominator addresses a different clinical question. First, all births will describe the chance of an infant to survive delivery. Second, all live births will calculate the probability that a baby born alive will survive initial care or resuscitation. Third, the denominator of all NICU admissions will describe the probability that an infant who was admitted to the NICU will leave the hospital. Fourth, the denominator 'infants discharged home' describes the longer-term survival chances once leaving the NICU (Guillen, DeMauro et al. 2011, Smith, Draper et al. 2014, Rysavy, Marlow et al. 2016). This means that survival outcomes can change from 18% among all live births to 60% among those admitted to NICU at 22 weeks' gestation. In this example from the population based cohort in Norway, three out of five babies – meaning 60% – survived after receiving active treatment (Stensvold, Klingenberg et al. 2017). Hence, outcome statistics, which have an important influence on decision-making, are substantially different depending on the denominator chosen. However, many publications still do not clearly report the denominator. To have a complete understanding of the reported data, each denominator should be clearly defined. For example, the

survival rate of delivery, the survival rate of resuscitation, and the survival rate of babies admitted to the NICU. Some scholars suggest to use the survival statistics of all dominators to inform parents of the likely survival of their baby depending on a specific decision-making time-point (Guillen, DeMauro et al. 2011).

Second, outcomes are either reported for a geographically defined population (i.e. a nationwide or regional study) or for a single hospital or set of hospitals. Closely related to the denominator bias described above, whether a study is population-based or hospital-based can affect the way that outcomes are reported. Most often, population-based studies report the outcome of all babies, within a given geographical area, alive at onset of labour, including stillborn infants at birth. Therefore, the rates of survival will be generally lower in comparison to hospital-based studies that report the outcomes of live births (i.e. live-born infants) or only neonatal unit admissions (Smith, Draper et al. 2014). Hence, it is important that a study's source population, whether single-centre, multicentre, or geographically defined, is clearly specified (Rysavy, Marlow et al. 2016).

Third, these differences in survival rates illustrate a selective use of perinatal interventions before 25 weeks of GA. Countries such as France, Switzerland, the Netherlands are known to limit care for babies born before 24 or even 25 weeks of gestation (Guillen, Weiss et al. 2015, Diguisto, Goffinet et al. 2017). Active obstetrical care (i.e. antenatal corticosteroids, antenatal transfer and caesarean section for foetal indication) or active life-sustaining measures in the delivery room (i.e. resuscitation and initiation of IC care) increase the chances of both survival and survival without morbidity for a baby born at this gestation (Kollee, Cuttini et al. 2009, Rysavy, Li et al. 2015). Withholding aforementioned active care may partly explain low survival rates. This points towards the issue of threshold policies (also known as gestational age policies), which can affect the mortality of babies born before 25 weeks of GA when they are considered to be at the threshold of viability and not eligible for intensive care treatment (for the Swiss recommendations see Box 1 below).

Box 1. Swiss Recommendations for Perinatal Care at the Limit of Viability (2011)

In Switzerland, the national recommendation maintains that treatment for babies born ≤23 weeks GA is not indicated and parents cannot insist on an unreasonable intervention. For babies born ≥25 weeks' treatment is recommended, consequently, parents cannot reject such interventions that are considered to be in the best interest of the infant.

- IC for babies born 23 0/7 – 6/7 is not indicated except for special situations such as the indication of several positive risk factors (e.g. estimated fetal weight, sex, antenatal corticosteroids, single or multiple birth). Then, provisional IC treatment may be initiated.
- Treatment for babies born 24 0/7 – 6/7 is conditionally recommended. Likewise, (positive or negative) risk factors can influence the decision. In such cases, the balance between long-term benefits of LSIs versus acceptability of the imposed burden are discussed with parents.

Hence, the formal grey zone is defined as 24 0/7 to 24 6/7, but both the upper and the lower limits can be extended into week 23 or into week 25, depending on the individual prognosis. Additionally, it is important to note that the Swiss recommendation recognizes the limitations of applying solely gestational age as a criterion for initiating IC. The recommendation states that 'apart from stratification by gestational age, factors significantly affecting prognosis must be taken into account' (Berger, Bernet et al. 2011).

A study from Norway, which shows the survival rates using all three denominators (births, live births and admissions to the NICU, see Table 2), reported that more infants born at 22 and 23 weeks were stillborn *after* being alive on intrauterine admission to an obstetrical unit (Stensvold, Klingenberg et al. 2017). This can indicate that a part of survival rates at this gestational age is influenced by the obstetrical decision to not actively intervene. This becomes problematic once such outcome data – for individual infants in whom intensive care treatment was withheld – is used to infer the probability of a good outcome for a birth where initiation of care is intended. This, then, skews the survival data. Of course, if no infants at, for example, 22 weeks GA were resuscitated,

no infants will survive at 22 weeks. When such low survival rates are the foundation of recommendations to withhold resuscitation for infants, this, in turn, creates a self-fulfilling prophecy (Wilkinson 2009, Brunkhorst, Weiner et al. 2014). This issue is particularly relevant for births at 22, 23, and 24 weeks' gestation, for which the provision of active treatment is most variable (Rysavy, Marlow et al. 2016). Hence, the relevant question, then, is not 'what are the overall survival statistics for this gestational age, but rather what would be the chance of survival and intact survival if maximal efforts were made' (Mercurio 2011). Again, this makes it important to report not only a study's source population or the denominator for survival rates, but also threshold policies.

Overall, an important limitation of population statistics is that outcome results cannot predict the individual outcome for a specific infant. If, in general, 30% of infants survive at 23 weeks of GA, there is no way to predict whether a specific infant falls within the 30% of survivors or within the 70% of non-survivors (Brunkhorst, Weiner et al. 2014). In fact, it is difficult to predict, before or at birth, which babies will survive, which will survive with impairments (severe or moderate), which will die, and which will survive unscathed (Lantos and Meadow 2006). Such prognostic uncertainty complicates decisions whether to initiate, withhold or withdraw intensive care treatment at birth or after admission into the NICU.

The realities of neonatal intensive care have shown that for extremely premature babies there is a very wide range of possible outcomes. But, several international studies have shown that not only *outcome data* vary widely but clinical cases are also *approached* differently in various countries (Cuttini, Casotto et al. 2006, Gallagher, Martin et al. 2014). These aspects will be addressed in the next paragraph.

The Attitudes and Values of Stakeholders

Accurate outcome data for the assessment of the individual infant are required for sound ethical reasoning to reach a clinical decision with the health care team and parents. Another aspect, however, frequently intervenes in the end-of-life decision-making process, namely the attitudes and values of the involved parties such as health care professionals and parents.

The first large cohort on neonatal end-of-life decision-making in Europe in the mid-90s showed that attitudes and values underlie end-of-life decisions. Health care professionals' age, length of professional experience and knowledge of follow-up of former preterm babies were shown to have a significant effect on the decision (Cuttini, Nadai et al. 2000). However, decisions

did not only rely on the characteristics of individual physicians or units, it was shown to be depended on the country and its culture as well. Cultural and country-specific factors were a strong predictor of physicians' personal attitudes and practices. This indicates that cultural and societal factors strongly affect practice regarding end-of-life decisions of extreme preterm infants (Rebagliato, Cuttini et al. 2000). A recent study from the Netherlands showed that attitudes towards initiating active treatment varied widely between individual perinatal professionals (Geurtzen, Draaisma et al. 2016). Active antenatal care in France is also shown to vary widely depending on local protocols and attitudes to management of extremely premature birth (Diguisto, Goffinet et al. 2017). A vast amount of studies have showed that resuscitation decisions of health care professionals (from neonatology, obstetrics and paediatrics) are based on many subjective factors. Often, preterm infants are denied intensive care treatment, compared with older patients whose outcomes are the same or worse (Janvier, Leblanc et al. 2008, Janvier, Leblanc et al. 2008, Armstrong, Ryan et al. 2011, Laventhal, Spelke et al. 2011, Hagen, Therkelsen et al. 2012, Hansen, Janvier et al. 2013, Mills, Janvier et al. 2015, Laventhal, Verhagen et al. 2016). HCPs mentioned considerations of personhood, a history of personal attachment and the family's interests as justifications for withholding care and providing comfort care to a baby born extremely premature, regardless of estimations that resuscitation was in the infants' best interests (Cuttini, Nadai et al. 2000, Marcello, Stefano et al. 2011, Cornfield and Kahn 2012, Dupont-Thibodeau, Hindie et al. 2017). Neonatal HCPs seem determined to avoid polarization of conflict and seek to find a compromise between the families and infants' best interest. In practice, this meant that caregivers were willing to give the final decision to parents (Moratti 2010). These different ways of understanding best interests suggests that the concept has 'little to do with survival and level of disability but more to do with other factors' (Mills, Janvier et al. 2015).

Accordingly, attitudes can influence or contribute to variations in survival and health outcomes, which can lead to wide variations in practice. This has already been pointed out in the previous paragraph on threshold policies. Such wide variations are, in part, due to different values, cultures and/or policy statements on caring for extremely premature infants. Outcomes as such are, then, determined by both the efficacy of the interventions as well as by underlying philosophies (Fischer, Steurer et al. 2009). Such an underlying philosophy of care for infants born extremely premature can differ between physicians, between hospital centres and between physicians working in different countries (Janvier, Nadeau et al. 2007). This is often called the 'NICU culture', where

a pattern of learned beliefs, values and behaviour that are shared within the group affect end-of-life decision-making (Berger 2010, Van McCrary, Green et al. 2014).

Hence, it is important to note that in the context of extreme prematurity there is an intimate relationship between values, policies, and facts and different policies and practices can make it difficult to interpret the data on neonatal outcomes (Verhagen and Janvier 2016). This illustrates the importance of being as explicit as possible in the communication with parents during end-of-life decision-making. On the one hand, explicitly take into consideration the research limitations affecting survival and long-term outcomes, and on the other hand, bear in mind that differences in attitudes and values can affect decisions at the end-of-life. Since the circumstances of death differ so widely between NICUs because of these diverging values and facts, we turn to the ethical reasoning regarding end-of-life decisions for extreme preterm infants.

End-of-Life Decision-Making for Infants Born at the Limit of Viability

Decisions in health care can be particularly difficult, involving a complex web of diagnostic and therapeutic uncertainties, patient preferences and values. Decisions regarding treatment for extreme preterm infants can be especially challenging since they rest on the dilemma that 'withholding or withdrawing treatment leads to certain death, yet initiating life-sustaining measures lead to an uncertain future with the possibilities ranging from death to considerable medical, emotional, societal, and financial risk to normal life' (Leuthner 2014).[3] Additionally, as shown in the previous paragraphs, good facts are necessary for ethical decision-making on neonatal intensive care for extreme preterm infants, 'but good facts do not necessarily lead to moral consensus. In neonatology, doctors, professional societies, bioethicists, parents, and judges may study the same data and come to fundamentally different conclusions' (Lantos and Meadow 2009). Hence, reasonable people can disagree whether to initiate or withhold intensive care measures or whether to continue or withdraw life-sustaining measures when there are uncertain outcomes. Disagreement also exists on who should be responsible for these decisions. So far, there is no consensus on these matters.

[3] It is important to note that end-of-life decisions to either withhold or withdraw care are considered ethically equivalent. It is permissible to withdraw treatment that a patient receives provided that it would have been permissible to withhold the same treatment (if it were not already being given) and vice versa (Larcher, Craig et al. 2015).

But without consensus one can still agree on the key ethical principles as well as the role of ethical analysis and the importance of a robust and an informed debate (Wilkinson and Savulescu 2017). Within this debate, it is important to focus on the ethical concepts (mid-level principles) that are essential to the concrete circumstances in neonatal medicine. It is, therefore, not necessary to settle on one normative framework (high-level theory) (Arras 2016). In the following paragraphs, I will introduce the main ethical concepts that are of importance in this debate which are the best interest principle, futility, quality of life, parental authority, the health care professionals' duty to care and shared decision-making.

The Best Interest Principle

Best interest is a beneficence-oriented ethical principle, which is widely considered crucial for making decisions for others. More specifically, the best interest principle plays a fundamental role in neonatology and is considered the prevailing standard (Leuthner 2014). Given that proxy decision-making for neonates cannot rely on the previous wishes or autonomous choices of the infant (the advance directives and substitute judgment principle), decisions rely instead on the best interest principle (Moro, Kavanaugh et al. 2006, Racine and Shevell 2009, Brock 2013). This involves selecting the treatment option that maximizes the patients', in this case the infants', overall good and minimizes the overall risks of harm (Brock 2013, Leuthner 2014). This means that the stakeholders involved in the decision consider what most reasonable persons would choose under such circumstances (Brock 2013). According to Buchanan and Brock, the best interest principle 'expresses a positive obligation, a duty to do what best promotes someone's interests or is most conducive to his or her good' (Buchanan and Brock 1989). Burdens can include the pain or discomfort from treatment and possible disabilities. Benefits can include the chance of survival and the pleasure the child may experience (short and long term) should he or she survive (Kopelman 2013). Under ideal circumstances, parents, together with health care professionals, weigh these benefits and burdens of different treatment options for the infant (Leuthner 2014).

Although the concept of best interest is central to ethical decision-making in neonatology, there are scholars who have pointed towards its allegedly controversial and vague features (Brody and Bartholome 1988, Veatch 1995, Salter 2012, Rhodes and Holzman 2014). The main problem with the best interest standard is that different people can value different things, they can prioritize differently and, therefore, reach different conclusions about what is best (Rhodes and Holzman

2014). Hence, controversy and possible conflict can arise when parents and HCPs view a child's best interests differently, based on their interpretation of what is best (i.e. different values) and assessment of the benefits/burdens (i.e. different understanding of relevant facts) (Leuthner 2014, Larcher, Craig et al. 2015 1580). I will highlight two elements that are commonly regarded as important in determining whether a decision to withhold or withdraw treatment can be in the child's best interest, namely futility and quality of life (Buchanan and Brock 1989).

Futile Care

Intensive-care medicine has become more and more confronted with ethical tensions between the demands of families and the appropriateness of certain interventions (Swiss Academy of Medical Sciences, Stocker et al. 2015). Central to decision-making is whether there is a limit to the care that a patient – or in this case parents – may demand. Differentiating between appropriate and inappropriate use of life-sustaining measures comes with numerous definitions of what constitutes as futile care (Brock 2013). All definitions (see Box 2 below) either refer to defining medical futility with odds (numbers/prognosis) or ends (observed ends that do not justify the efforts).

Box 2. Five Definitions of Futility

- *Physiological futility* covers treatment that does not produce a physiological response to treatment.
- *Imminent demise futility* is when death is to be expected and the patient is likely to die in the near future.
- *Lethal condition futility* considers care futile when a known illness will lead to near death.
- *Qualitative futility* defines care futile when no acceptable quality of life results from further treatment.
- *Quantitative futility* – the most cited definition – refers to (1) the very low probability (no more than 1%) of a treatment having effect, (2) treatment merely persevering unconsciousness, or (3) treatment fails to end dependence on intensive care treatment (Schneiderman, Jecker et al. 1990, Brody and Halevy 1995, Krones and Monteverde 2013).

The main problem of any definition of futility is setting the value of odds or ends. In fact, only a zero percent change of a treatment having effect would be the value representing 'pure futility' (Nair-Collins 2015). Consequently, depending on which definition is adopted life-sustaining

measures may or may not be considered appropriate. These judgments have an intrinsically moral nature and different values influence what counts as futile treatment. Some guidelines aim to avoid the ambiguity of the concept of futility by referring to treatment as ‘ineffective’ or with ‘little or no likelihood of benefit’. When the defined goal is not attained and no potential remediable causes exist, treatment is considered ineffective. Additionally, treatment offers little or no likelihood of benefit when there is no reasonable prospect of the patient being able to return to an appropriate living environment. In these two cases, treatment should be discontinued (Swiss Academy of Medical Sciences, Stocker et al. 2015). Despite the fact that there is no one-to-one overlap with the futility concepts, the ends are similar. If treatment cannot reach its therapeutic goal, the efforts cannot be justified. No odds are mentioned and the difficulty with this approach is that there is no absolute certainty on when treatment is considered illegitimate.

Although wide agreement exists that treatment is ‘futile’ or ‘ineffective’ if it does not reach its purpose or if it cannot benefit the patient, as of yet no agreement on what counts as non-beneficial treatment has been achieved (Krones and Monteverde 2013, Nair-Collins 2015). In turn, there are two main scenarios in making decisions for extremely premature infants. Sometimes life-sustaining measures are withheld or withdrawn from infants whose survival is extremely unlikely (quantitative futility or ineffective treatment). At other times, life-sustaining measures are withheld or withdrawn from infants who might survive if all treatment were provided (qualitative futility or little or no likelihood of benefit) (Krones and Monteverde 2013, Swiss Academy of Medical Sciences, Stocker et al. 2015). In such cases, decisions to withhold or withdraw are based upon predicted quality of life and are far more contentious (Wilkinson 2011, Verhagen and Janvier 2016). This, then, raises important ethical questions because decisions are no longer grounded in objective empirical facts, instead these decisions raise some discussions about quality of life which are inevitably moral discussions about what it means to have a live not worth living. In fact, these are the cases relevant to the ethical discussion that I will fall back on time and again throughout this thesis. In the next paragraph, the difficulties of assessing an infant’s future quality of life will be discussed.

Quality of Life

Clinical decision-making in neonatology aims to avoid the indefinite prolongation of life (the ‘quantity’ of life) and intends to ensure that the life that is prolonged meets certain criteria (of

'quality'). Consequently, decisions about withholding or withdrawing care are based on long-term projections of the child's future quality of life (QoL). QoL is a broad multidimensional concept with an evaluative meaning, which implies comparison and quantification of an individual life experience against an objective standard. The purpose of a QoL determination is to decide when a life lacks sufficient quality and it is ought not a live worth living. QoL is assessed across a series of domains such as material, physical, social, emotional and productive well-being (Wyatt 2011). One such tool is the health related quality of life (HRQoL), which combines objective assessments of functional ability with subjective reports on emotional well-being. HRQoL is used as a tool to measure the evolving perception of HRQoL throughout its development within the family. Studies have shown that the impact of being born preterm is greatest at younger age and ameliorates over time. HRQoL determinations at young ages show poorer health and lower QoL scores compared with peers born at term. What most studies clearly show is that having a physical or neurological impairment does not necessarily translate into a poor self-assessed quality of life (Saigal and Tyson 2008, Huhtala, Korja et al. 2016). Although there is ongoing debate on whether these positive self-assessments should be considered as denial or successful adjustment and coping mechanisms, there is general agreement that the perception of the affected individual is the most important.

According to Payot (2016) quality of life considerations raise two concerns. First, health care professionals, parents and survivors greatly differ in the evaluation of quality of life of an individual patient. Second, contextual factors (such as the families socio-economic background, education and support system) have a great influence on the outcome of premature babies impacting their future quality of life (Payot 2016). These factors can lead to situations where a diminished QoL shapes the boundaries of medical intervention. First, one of the main problems is that the HRQoL is often rated by parents or HCPs rather than by the children themselves. As a matter of fact, disabilities and well-being are more negatively rated by HCPs, and a little less so by parents, than by the children who were born extremely premature (Zwicker and Harris 2008). Given these facts, HCPs could face a conflict between their often negative prediction of an infant's long-term prognosis and the parents' (potential) desire for continued life-sustaining treatment (Janvier, Barrington et al. 2012). Another limitation in the evaluation of the QoL in babies born extremely premature is the fact that assessments are not measurements but rather predictions, which remain clouded by huge prognostic uncertainty. Such a prediction fails to take into account the nuances of living after being born extremely premature such as the child's (and family's) resiliency to overcome physical disabilities and develop social and relational skills. Accordingly, the

quantitative assessment of a child's future QoL may be very different from the child's future subjective experience of their QoL (Green, Darbyshire et al. 2016, Payot 2016). Another difficulty is that our values and preferences, hence our subjective evaluations of our own QoL, change as we grow older. One might even speak of family resilience – the parents' ability to adapt to the situation – in developing a nurturing environment in which a child with potential disabilities can grow up. On the contrary, in some situations parents' views may be negatively affected by the burden of caregiving, stress, quality of follow-up services and help provided (e.g. from the health care system or the family's social environment), and their own mental, social and economic status.

This raises the second issue with HRQoL assessments. Studies show that in general families of extreme preterm infants experience more stress, but also greater family cohesion. As previously shown, stress is often ameliorated over time; however, for some families this is not the case. Families with little social support do not experience a reduction of stress, even after many years (Singer, Fulton et al. 2010). Accordingly, it appears to be worse for an extremely premature baby to be born within a family with a low educational background or low socio-economic status. The neurodevelopmental trajectories in children from mothers with high educational background were more promising compared with those from less-educated mothers. Possible explanations may be quality and quantity of cognitive stimulation, the parenting style, both associated with educational background or genetics (Voss, Jungmann et al. 2012). This might be related to the availability and access of support systems for families with low socio-economic and educational backgrounds, making it especially important to provide these parents with special support and follow-up care services.

In summary, QoL requires careful judgments to be made that acknowledge the subjectivity of evaluations (i.e. depending on the stakeholder and time-period) and recognize the influence of socio-economic factors that may (in)directly influence the QoL in terms of services and help provided to individuals with impairments. There seems to be no easy answer to the questions on what life is worth being lived and when is death better than a life with disabilities (Janvier, Barrington et al. 2012). Moreover, QoL, futility and a best interest assessment cannot be considered as objective parameters. They each involve subjective and highly contestable value judgments (Inwald 2008, Wyatt 2011, Krones and Monteverde 2013).

Parental Authority and the Health Care Professionals' Duty to Care

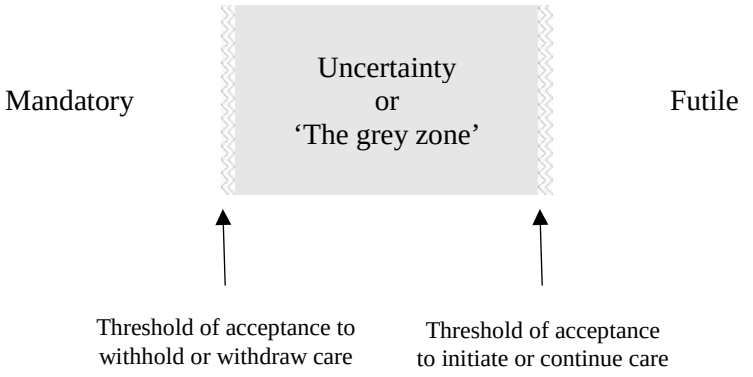
An additional challenging factor in the context of extreme prematurity is that these patients cannot make autonomous choices regarding their own care. In neonatal and paediatric cases, parents are considered to have the legal and moral authority to make medical decisions for their children. This is based on the conviction that parents are best able to ascertain their children's best interests and act in accordance (Leuthner 2014). In turn, health care professionals' have the moral obligation and duty to act in the best interest of their patients. Treatment should maximize benefits and minimize harms, consistent with its intended purpose and the goal of every therapy. Consequently, the health care team should introduce treatments that benefits the child and should consider withholding or withdrawing such treatments when there is no benefit, or when the burden is considered inacceptable. Besides the principle of respecting the life and health of patients by aiming to restore health and preventing disease, HCPs also have the duty to respect parental rights and priorities, and make sure that parents have enough information to make a thoughtful decision together with the health care team (Unguru 2011, Larcher, Craig et al. 2015). Until now no consensus is found on who should be responsible for these decisions, i.e. should parents, health care professionals or the state have the final responsibility and say in these decisions (Lantos and Meadow 2006, Dupont-Thibodeau, Barrington et al. 2014). In France, for example, no formal consent of the parents is needed for decisions to withhold or withdraw care, since the authority of health care professionals overtakes parental authority. Whereas in the United States parental authority is central in decision-making for extreme preterm infants (Orfali 2004, Sauer, Dorscheidt et al. 2013).

Although it is often possible for neonatal HCPs and parents to find an agreement whether to continue, to withhold or withdraw IC treatment on best interest grounds, in some rare cases conflict can arise. In essence, there are two circumstances where HCPs and parents may disagree. First, the parents may wish limitation of treatments that the health care team regards as being in the infant's best interest. Second, HCPs may consider withholding or withdrawing treatment as in the infant's best interest, but parents disagree and want to continue IC treatment (Larcher, Craig et al. 2015). Such disagreements are either the result of diverging *interpretations* of what is in the infants' best interest between HCPs and parents – due to different values or a different understanding of the relevant facts – or the families' and the infant's interests diverge. In cases where parental preferences might harm the child, the HCP must guard the welfare of the patient and protect their interests. This raises the dilemma of limiting parental authority to act on what is in the best interest

of the infant with the physicians' duty of care for critically ill newborns (Gomez, Bielefeld et al. 2016). However, some critiques have been raised on the difficulty of identifying the best interest of the infant and the limits set to parental authority (Unguru 2011, Larcher, Craig et al. 2015).

One of the main critiques to a best interest assessment is that the principle solely focuses on the infant's interests and negates the interests of others. Hence, the best interest of the infant stands in opposition with the interests of the family, as if they are not related. This appears to conflict, then, with the major role of parents in decisions about life-sustaining treatment in most parts of the world (Wilkinson 2013). In light of these concerns, three alternate approaches have been proposed. The concept of ***constrained parental autonomy*** focuses on the infant's basic needs and not his or her best interest. As a consequence, this concept does not comprise the needs of the family. Parents are granted autonomy to make decisions with an emphasis on the family's interests by accommodating 'intra-familial trade-offs provided that the basic needs of each child member are secured' (Ross 1998). The ***harm principle*** ensures that health care decisions do not cause significant harm to the patient. Similar to the constrained parental autonomy concept, the harm principle allows for more flexibility to parental decision-making and, in turn, generates a threshold for harm (Diekema 2004). The ***threshold framework*** argues for an upper and lower limit to parental discretion. These thresholds move alongside two extremes. On the one hand, prognosis can be so good that treatment must be provided, even if parents disagree. This is the threshold of mandatory treatment or the upper threshold. On the other hand, prognosis can be so poor that life-sustaining measures should be limited, even if parents request (continuation of) IC treatment. This is the threshold of inappropriate treatment or the lower threshold (Wilkinson 2013). These thresholds relate to the concept mentioned earlier (see Figure 3), where extreme preterm infants are classified into three groups for which care is either mandatory, optional or unreasonable (Tyson 1995). The problem of defining the boundaries of these thresholds and the arbitrariness of limiting life-sustaining measures before or at birth for specific groups of extreme preterm infants will be discussed in-depth in Part III in the Chapter *'Fragile lives with Fragile Rights'*.

Figure 3. Three different categories that affect end-of-life decisions



However, if one allows the family's interest and values to define what is best then it seems subjective (Leuthner 2014). A parental decision can be determined by the families own system of values such as religious beliefs, cultural attitudes or parents lived experiences (Larcher, Craig et al. 2015). But an approach that aims to include the family's interests does not ignore the interests of the child, nor does it allow parents to have the final say in all decisions relating to their child. These approaches recognize that the interests of an infant are difficult to separate entirely from the interests of the family and that in end-of-life decisions the quality of life of that child can only be properly understood in the context of the family (Inwald 2008). Furthermore, we should not forget that when HCPs focus 'solely on the infant's best interests' this judgement lies exclusively with the health care team, which can be affected by their personal values and attitudes and is open to bias as well (McHaffie, Laing et al. 2001, Frost, Cook et al. 2011, Wilkinson and Truog 2013).

One way to balance different best interest perspectives and to approach the identification of the child's best interests is to rely on a shared decision-making model. The fundamental goal of shared decision-making is to benefit from and include the HCPs' and the parents' respective knowledge and experience in the decision-making process (Racine and Shevell 2009).

Shared Decision-Making

Traditionally, health care professional would make decisions for extreme preterm infants without discussing it with parents. This paternalistic tradition of 'the doctor knows best' has changed during the 20th century toward a shared decision-making approach (Charles, Gafni et al. 1997). Currently, most professional guidelines recommend that HCPs share decision-making authority with parents of babies born premature (Nuffield Council on Bioethics 2006). This means that HCPs and parents act as partners to promote the best interests of the child (Leuthner 2001,

Racine and Shevell 2009, Larcher, Craig et al. 2015). In this sense, the HCPs' medical expertise guides the family in the decision-making process. This is an interdependent process between HCPs and parents who collaborate in making decisions about the infant's health care. Shared decision-making, thus, integrates the medical evidence and the provider's clinical expertise about each individual child (i.e. patient specific) and relies on the unique attributes of the patient and his or her family (i.e. their values and interests) (Charles, Gafni et al. 1997, Stiggelbout, Van der Weijden et al. 2012, Légaré and Witteman 2013).

At the heart of shared decision-making lies proactive communication and empowerment of parents. Communication should be as clear and honest as possible to enhance trust and confidence and to avoid conflicts. Furthermore, parents should be empowered by HCPs to be involved in the care of their child and to clarify their value priorities. This way HCPs provide the medical knowledge regarding the expected prognosis and response to medical treatment, while parents provide the moral knowledge of the benefits and burden and give meaning to the predicted outcome. This implies that parental values influence what actions are in an infant's best interests (Leuthner 2001). This further means that parents should receive clear, unbiased, understandable, and complete information to consider all possible therapeutic options, understand the risks, benefits and possible outcomes. This, in turn, enables parents to fully participate, should they want to, in the decision for the infant's best interest that is according to their cultural, religious, economic and ethical values (D'Aloja, Floris et al. 2010).

Based on the previous paragraphs we know that in some situations decisions are made not on the basis of prognosis but on the *meaning* of that prognosis. Hence, best interest decisions include a combination of medical and moral values. In cases where parents and HCPs do not agree on the best treatment for the extreme preterm infants, deliberation should be sought in line with the best shareable choice for the infant. It is important to recognize the prognostic and moral uncertainty in caring for extreme preterm babies, best interest is not objective, quality of life evaluations are individualistic, and futile care is difficult to define. When faced with such uncertainty and a range of different attitudes and values shared decision-making as a collaborative relationship between HCPs and parents is key to negotiate the meaning of an infant's best interest (Leuthner 2001). Negotiation thus means more than respecting parents' rights in decision-making. It means involving parents in the process of decision-making (Mahgoub, van Manen et al. 2014). The role

of shared decision-making in negotiating and assessing the best interest of the infant will be further discussed in the final chapter.

Research Objectives

Against this epidemiological and theoretical background, this study aims at exploring the actual societal attitudes and parents' experiences with regard to end-of-life decision-making and withholding or withdrawing treatment in extremely premature infants in Switzerland. It concludes with a theoretical reflection on whether we should offer life-sustaining intensive care measures to all parents of babies born extremely premature. This thesis consists of three parts.

In Part I, I present the results from the population survey on the societal attitudes and values regarding extreme prematurity in Switzerland. To date, empirical data have provided limited insight into the effect of societal attitudes and values on end-of-life decision-making for infants born at the threshold of viability in order to foster a transparent discussion on attitudes and values for reconciling medical and ethical deliberations with socio-religious and personal predispositions in end-of-life decisions. This study further aims to explore whether socio-cultural differences would lead to diverging end-of-life decisions and touches upon the question of solidarity (as the willingness to carry the costs) for extremely preterm infants at risk for disabilities.

In Part II, I explore the experiences of parents who lost an extremely premature baby in the delivery room or in the neonatal intensive care unit (NICU) of the University Hospital Zurich. Still little is known in practice about parental experiences with end-of-life decisions. Particularly in Switzerland, the legal, ethical, and societal debates take place with scant knowledge and information on the actual attitudes and values of parents that underlie end-of-life decisions in extremely preterm infants. Moreover, this study investigates how parents perceive communication, information and care in this process and which aspects in terms of decision-making and support parents found particularly helpful or challenging. I will furthermore illustrate experiences of parental bonding with an extremely premature baby and end-of-life decision-making.

In Part III, I present a theoretical reflection on policies that limit intensive care measures for babies at a certain gestational age. This study component explores the reasons why many professional societies or hospitals, throughout Europe and North America, remain to have such policies regarding the treatment of extremely premature babies. It furthermore questions its ethical justifications.

In the general discussion, the findings of the three studies are summarized, strengths and limitations are discussed, and implications for policy and future research are suggested.

PART I SOCIETAL ATTITUDES AND VALUES

Attitudes towards Decisions about Extremely Premature Infants

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Author Contributions

All authors have contributed to the different parts of the manuscript, and each author has participated sufficiently in the work to take public responsibility for appropriate portions of the content. Miss Hendriks and Dr. Klein carried out the data extraction and analysis, interpreted the data, wrote the initial manuscript, and approved the final manuscript as submitted. Drs. Bucher and Fauchère designed the study, drafted the questionnaire, interpreted the data, critically reviewed and revised the manuscript, and approved the final manuscript as submitted. Dr. Streuli interpreted the data, wrote the initial manuscript, and approved the final manuscript as submitted. Dr. Baumann-Hölzle designed the study, interpreted the data, critically reviewed and revised the manuscript, and approved the final manuscript as submitted. All authors have read and approved the submission of the manuscript and take full responsibility.

Hendriks, M. J., et. al. (2017), Attitudes towards decisions about extremely premature infants differed between Swiss linguistic regions in population-based study. *Acta Paediatr*, 106: 423–429. doi:10.1111/apa.13680

Abstract

Aim: Studies have provided insights into the different attitudes and values of healthcare professionals and parents toward extreme prematurity. This study explored societal attitudes and values in Switzerland with regard to this patient group.

Methods: A nationwide trilingual telephone survey was conducted in the French, German and Italian speaking regions of Switzerland to explore the general population's attitudes and values with regard to extreme prematurity. Swiss residents of 18 years or older were recruited from the official telephone registry using quota sampling and a logistic regression model assessed the influence of socio-demographic factors on end-of-life decision-making.

Results: Of the 5,112 people contacted, 1,210 (23.7%) participated. Of these 5% were the parents of a premature infant and 26% knew parents with a premature infant. Most participants (77.8%) highlighted their strong preference for shared decision-making and 64.6% said that if there was dissent then the parents should have the final word. Overall, our logistic regression model showed that regional differences were the most significant factors influencing decision-making.

Conclusion: The majority of the Swiss population clearly favoured shared decision-making. The context of socio-cultural demographics, especially the linguistic region in which the decision-making took place, strongly influenced attitudes toward extreme prematurity and decision-making.

Keywords: End-of-life decision making, extreme prematurity, moral pluralism, population survey, shared decision-making

Key Notes

- A nationwide telephone survey was conducted in the French, German and Italian speaking regions of Switzerland to explore the general population's attitudes and values with regard to extreme prematurity.
- Most of the 1,210 adults (77.8%) highlighted their strong preference for shared decision-making and 64.6% said parents should have the final word in cases of dissent.
- Our logistic regression model showed that regional differences were the most significant factors influencing decision-making.

Introduction

Despite a steady increase in survival rates for infants born at the limits of viability (Seaton, King et al. 2013, Serenius, Sjors et al. 2014), long-term morbidity has not decreased accordingly. Data on long-term outcome and quality of life (QoL) are essential for decision-making, but their interpretations vary largely (Tommiska, Heinonen et al. 2007, Anderson, Baer et al. 2016, Holsti, Adamsson et al. 2016).

This prognostic uncertainty raises various difficult ethical questions. For instance, decision-making can be influenced by the diverging attitudes and values of healthcare professionals, parents and society at large with regard to treatment options for infants born extremely premature. Diverging attitudes have led to considerable variability in clinical practice, predominantly caused by cultural differences and personal beliefs (Garel, Caeymaex et al. 2011, Fanaroff, Hascoet et al. 2014, Gallagher, Martin et al. 2014). Also, the degree of parental involvement in medical decision-making for extremely preterm infants has shown to be embedded in the diverging social and cultural practices in France, the United States and Canada (Orfali 2004, Marcello, Stefano et al. 2011).

A transparent discussion on attitudes and values is necessary to reconcile medical and ethical deliberations with socio-religious and personal predispositions in end-of-life (EoL) decisions. Assessing the views and values of a given society may be helpful in understanding the opinions and views put forward by parents and neonatal staff and can illustrate how disagreements about end-of-life decision-making (EoLDM) can arise.

To date, empirical data have provided limited insight into the effect of societal attitudes and values on EoLDM for infants born at the threshold of viability. Therefore, the purpose of this study was to examine the attitudes and values within the Swiss population toward extreme prematurity, in order to assess whether socio-cultural or religious-based differences would lead to diverging EoL decisions.

Methods

Design and population

For this population-based telephone survey, the initial German questionnaire was translated into French and Italian. The translation accuracy was checked with back-translation into German and reviewed by a panel of translators to ensure identical semantic content in each language.

The questionnaire was used to assess the participant's opinions, attitudes and views regarding extremely preterm infants. Participants were informed that extremely preterm infants were those born before 28 weeks of gestation, with a birth weight of between 600 and 800 grams, who always required intensive care treatment.

The survey consisted of 16 questions with a mean interview duration of 13.8 minutes and a 95% confidence interval (95% CI) of 13.5 to 14.0 minutes. This paper focuses on the following themes: decision-making about intensive care measures, views on intensive care measures and associations with acceptable QoL. Furthermore, demographic information was gathered on gender, age, residency, importance of religion, education and income level, personal experience with prematurity and whether the participants had children. This included whether they had premature children themselves or knew parents who had.

Data collection

The nationwide telephone survey was conducted in 2013 by the market and social research institute gfs-zurich, which recruited adults of 18 years or older living in Switzerland from the official telephone registry by means of quota sampling. Quotas were set in terms of sex, age and residential location for each linguistic region to make our sample representative of the demographic distribution of Switzerland.

As this anonymous population survey did not include any patient data or health-related data, the Ethical Committee of the Canton of Zurich did not require formal ethical approval.

Statistical analysis

Only 4% of the Swiss population live in the Italian-speaking part. 200 of the 1,210 interviewees were selected from the Italian-speaking region to obtain a larger sample to allow for comparisons between the three language areas. For comparisons between groups, for example levels of education and importance of religion, the answers were weighted so that respondents from the Italian-speaking region accounted for 4% of the answers. The weighing process was based on the official data of the Federal Statistical Office and allowed to get a precise sociodemographic picture of the Swiss population. For proportions, 95% CIs were calculated according to the formula devised by Wilson and reported where appropriate. Groups were compared using a chi-square test.

A logistic regression model was performed to assess the association between medical decision-making and socio-demographic factors. Factors included linguistic region, educational level,

gender and strength of faith and were chosen because of their significance in a preliminary univariate analysis. Odds ratios and 95% CIs were calculated from single factors of the logit function and a p value of < 0.05 was considered statistically significant.

Weighting and analysis were conducted using IBM SPSS Statistics software, version 22 (IBM Corp, New York, USA).

Results

Sample

Of the 5,112 people contacted, 1,210 completed the telephone survey, giving us a response rate of 23.7%. The main characteristics of the respondents are shown in Table 1. One in 20 of the respondents (5%) was the parent of a premature infant and 26% knew the parents of a preterm infant in their close social environment. Religious faith was stronger in the German-speaking (54.1%) and Italian-speaking region (53.3%) than in the French-speaking area (41.6%).

Table 1. Characteristics of respondents in the survey

Characteristic	%	N
Gender		
Male	49.7	602
Female	50.3	608
Region[a]		
German part	72.0	707
French part	24.0	303
Italian part	4.0	200
Age		
≤ 39 years	35.0	423
40-64 years	45.0	544
≥ 65 years	20.1	243
Having own children		
Yes	70.2	837
No	29.8	373

Characteristic	%	N
Personal experience with prematurity		
Weak	68.2	831
Strong	31.5	375
Importance of religion		
Not important	48.9	586
Important	51.1	606
Education		
Low	7.2	99
Medium	66.1	805
High	26.6	297

Total N=1210. Percentage without missing values unless indicated.

[a] Regions are based on: the German speaking part, the French speaking part and the Italian speaking part of Switzerland. All results in % are weighted for regions; numbers of respondents (N) are unweighted.

Decision-Making about Intensive Care Treatment Options

In terms of intensive care treatment decisions, the majority (77.8%) of the population preferred shared decisions between parents and healthcare professionals (Table 2), as the decision-making model for infants born extremely premature. Few people (2.6%) would confer the decisional authority to the ethics committee. Socio-demographic factors that had a significant effect on responses were age and linguistic region. People under 39 years of age were more likely (82.7%) to regard shared decision-making as important than older respondents of 40-64 years (75.6%) and 65 years or older (74.4%). Furthermore, giving physicians the sole authority to make decisions was higher in the Italian-speaking (10.5%) than German-speaking (2.5%) and French-speaking (2.6%) regions. The healthcare team had more authority in the Italian-speaking (8.0%) and French-speaking (6.3%) regions than the German-speaking area (2.0%).

Table 2a. Medical decision-making

Who should decide about using intensive care	All N % (95% CI)	≤ 39 years N % (95% CI)	40-64 years N % (95% CI)	≥ 65 years N % (95% CI)	Age p value	German N % (95% CI)	French N % (95% CI)	Italian N % (95% CI)	Region p value
The physician	47 2.9 (2.1-4.0)	10 1.9 (1.0-3.7)	22 3.3 (2.1-5.2)	15 4.1 (2.3-7.4)	0.217	18 2.5 (1.6-4.0)	8 2.6 (1.3-5.1)	21 10.5 (7.0-15.5)	**<0.001**
Health care team	49 3.2 (2.4-4.4)	13 2.6 (1.5-4.6)	25 3.7 (2.4-5.6)	11 3.3 (1.7-6.4)	0.641	14 2.0 (1.2-3.3)	19 6.3 (4.1-9.6)	16 8.0 (5.0-12.6)	**<0.001**
Parents	114 9.8 (8.2-11.6)	34 7.8 (5.6-10.8)	56 11.0 (8.7-13.9)	24 10.3 (7.1-14.8)	0.232	80 11.3 (9.2-13.9)	16 5.3 (3.3-8.4)	18 9.0 (5.8-13.8)	**0.011**
Parents and health care team	923 77.8	345 82.7 (78.9-86.0)	406 75.6 (71.8-79.0)	172 74.4 (68.5-79.5)	**0.010**	550 77.8 (74.6-80.7)	241 79.5 (74.6-83.7)	132 66.0 (59.2-72.2)	**0.001**

Who should decide about using intensive care	All N % (95% CI)	≤ 39 years N % (95% CI)	40-64 years N % (95% CI)	≥ 65 years N % (95% CI)	Age p value	German N % (95% CI)	French N % (95% CI)	Italian N % (95% CI)	Region p value
	(75.3-80.0)								
Ethics committee	35 2.6 (1.8-3.6)	9 1.9 (1.0-3.7)	22 3.9 (2.5-5.8)	4 0.8 (0.2-3.0)	**0.025**	16 2.3 (1.4-3.6)	10 3.3 (1.8-6.0)	9 4.5 (2.4-8.3)	0.221
Others[a]	31 2.9 (2.1-4.0)	6 1.7 (0.8-3.4)	11 2.2 (1.3-3.8)	14 6.2 (3.8-10.0)	**0.002**	23 3.3 (2.2-4.8)	6 2.0 (0.9-4.3)	2 1.0 (0.3-3.6)	0.156
Do not know	11 0.9 (0.5-1.6)	6 1.4 (0.7-3.1)	2 0.4 (0.1-1.3)	3 0.8 (0.2-3.0)	0.202	6 0.8 (0.4-1.8)	3 1.0 (0.3-2.9)	2 1.0 (0.3-3.6)	0.966

Table 2b. Medical decision-making

In case of disagreement, who should have the final say	All N % (95% CI)	≤ 39 years N % (95% CI)	40-64 years N % (95% CI)	≥ 65 years N % (95% CI)	Age p value	German N % (95% CI)	French N % (95% CI)	Italian N % (95% CI)	Region p value
The physician	163 12.3 (10.6-14.3)	48 9.7 (7.2-12.9)	75 13.1 (10.5-16.1)	40 15.6 (11.6-20.7)	0.069	72 10.2 (8.2-12.6)	54 17.8 (13.9-22.5)	37 18.5 (13.7-24.5)	**<0.001**
Health care team	40 3.4 (2.5-4.6)	11 2.8 (1.6-4.9)	19 3.5 (2.2-5.4)	10 4.1 (2.2-7.4)	0.675	22 3.1 (2.1-4.7)	13 4.3 (2.5-7.2)	5 2.5 (1.1-5.7)	0.494
Parents	756 64.6 (61.9-67.3)	281 70.0 (65.4-74.1)	337 63.4 (59.3-67.4)	138 57.8 (51.5-63.8)	**0.005**	494 69.9 (66.4-73.1)	154 50.8 (45.2-56.4)	108 54.0 (47.1-60.8)	**<0.001**

In case of disagreement, who should have the final say	All N % (95% CI)	≤ 39 years N % (95% CI)	40-64 years N % (95% CI)	≥ 65 years N % (95% CI)	Age p value	German N % (95% CI)	French N % (95% CI)	Italian N % (95% CI)	Region p value
Ethics committee	150 11.4 (9.7-13.3)	54 11.6 (8.9-15.0)	69 11.9 (9.5-14.9)	27 9.8 (6.7-14.2)	0.681	66 9.3 (7.4-11.7)	51 16.8 (13.0-21.5)	33 16.5 (12.0-22.3)	**<0.001**
Others[a]	36 3.2 (2.4-4.4)	3 0.7 (0.2-2.1)	17 3.3 (2.1-5.2)	16 7.4 (4.7-11.4)	**<0.001**	21 3.0 (2.0-4.5)	13 4.3 (2.5-7.2)	2 1.0 (0.3-3.6)	0.104
Do not know	65 5.0 (3.9-6.3)	26 5.2 (3.5-7.7)	27 4.8 (3.3-6.9)	12 5.3 (3.1-8.9)	0.931	32 4.5 (3.2-6.3)	18 5.9 (3.8-9.2)	15 7.5 (4.6-12.0)	0.227

In cases where there was dissent between the parents and the healthcare team, 64.6% stated that parents should have the final word in decision-making. Only 11.4% of the respondents specified the ethics committee as the final decision-making authority. Again, the socio-demographic factors age and linguistic area shaped the opinions on decision-making (Table 2). Compared to younger people under 40 years of age (70.0%), older people over 65 years (57.8%) were less likely to say that the parents should be the final decision-makers. In the German-speaking region, parents were more often (69.9%) designated as the final decision-makers. Respondents from the French-speaking (17.8%) and Italian-speaking areas (18.5%) gave more final power to the physician to make decisions than the German-speaking region (10.2%).

Research showed that the criteria for the initiation of intensive care treatment were often stricter for extremely preterm infants than for older children with the same prognosis for long-term outcome (Prentice, Janvier et al. 2016). The majority (62.6%) of participants felt the criteria for intensive care treatment should be the same, 17.8% said it should be stricter and 12.7% said the criteria should be less strict. Male respondents and those who were childless or very religious advocated more liberal criteria for initiating intensive care in extremely preterm infants.

Perceptions of QoL Aspects

There were a number of different responses to the open question about the criteria for an acceptable QoL for infants born extremely preterm. The four most common answers to this question were: being able to have an independent life (32%), being like others (18%), living without medical assistance (10%) or living without a disability (8%) and 6% of respondents could not answer the question. The minimum criteria for an acceptable QoL, with multiple responses possible, included: showing happiness and emotions (94.4%), communication with others (93.6%), taking care of oneself (75.7%), completing primary school (73.0%) and earning one‘s own living (65.2%).

Irrespective of the assumed future QoL, 45.2% believed that all possible therapeutic measures should be undertaken to enable the survival of extremely preterm infants, while 43.1% did not agree, and 11.7% did not know. It was significant that fewer French-speaking than German-speaking or Italian-speaking people considered all possible measures should be undertaken. Female respondents, people with a lower education or with strong religion supported intensive care treatment regardless of QoL considerations (Table 3).

Table 3. Results of multivariate logistic regression [a]

Characteristics	OR (95% CI)	p
Region		
German part	1	
French part	0.59 (0.43-0.80)	**0.001**
Italian part	1.98 (0.95-4.13)	0.067
Education		
Low	2.49 (1.45-4.27)	**0.001**
Intermediate	1	
High	1.02 (0.77-1.35)	0.901
Gender		
Male	1	
Female	1.36 (1.06-1.76)	**0.016**
Importance of Religion		
Not important	1	
Important	1.41 (1.10-1.81)	**0.008**

[a] Question: "Do you think that for extremely preterm infants, irrespective of the assumed future quality of life, all possible therapeutic measures should be undertaken to enable their survival? (yes/no)"
Higher ORs indicates higher agreement with the statement.

Discussion

This representative Swiss population-based study aimed to provide information about the current knowledge of lay-people on complex questions regarding EoL decisions for infants born extremely premature. This survey found distinct preferences regarding the type of decision-making and also demonstrated significant socio-cultural differences in the EoLDM process.

Our study found that the Swiss population explicitly preferred to reach consensus on an EoL decision through a shared dialogue between the healthcare team and parents. This is in contrast to

a paternalistic approach where decisions are solely made by the healthcare team or by the physician. Such a collaborative approach allows healthcare staff to take into account social and familial factors at play in the decision-making process. This preference confirms the previously reported importance of informed and shared decisions in paediatrics (de Vos, Bos et al. 2015) and, more specifically, in EoL decisions for infants born at the limit of viability (Berger, Bernet et al. 2011, Brunkhorst, Weiner et al. 2014). In cases where there were diverging views between parents and the healthcare team, people clearly favoured an approach in which the parents acted as the final decision-makers. This was consistent with other studies that considered parental authority as imperative in the final decision (de Vos, Bos et al. , Sullivan, Gillam et al. 2015), although implementation of parental authority still varies extensively (Pignotti and Donzelli 2015). Interestingly, very few of the Swiss people in our study said that they would rely on the expertise of an independent ethical committee with regard to EoL decisions, even in situations of dissent, while many other countries rely strongly on ethical committees to mediate in problematic decision-making issues (Mercurio 2011).

Despite the increasing importance of parental authority in cases of disagreement, in clinical practice it can be unclear how to balance familial autonomy with the best interest of the child. An in-depth discussion on parental autonomy is beyond the scope of this paper, but it is important to contextualise surrogate decision-making. This notion supports the principle of respecting the patients' autonomy, which in the context of neonates refers to the parental autonomy as the legal proxy of the child. The Swiss Civil Code for the protections of adults and children, enacted in 2013, clearly promotes and re-emphasises parental authority and family autonomy when a paediatric patient is incapable of taking his or her own decisions (Schweizerisches Zivilgesetzbuch 2008/2013). Since decision-making by proxy cannot be based on the prior wishes, views and values of the infant, it is therefore based on the best interests and assumed future QoL of the child (Rebagliato, Cuttini et al. 2000). Although healthcare teams in neonatal intensive care units (NICUs) aim to provide extremely premature infants with the best QoL possible, this term can be interpreted in many ways. Our study revealed that the overall perceptions of the population seemed related to future considerations and notions such as independence. This pattern has also been identified by other surveys on physicians' assessments of QoL in which the child's predicted suffering, inability to communicate and future autonomy were given increasing importance (Verhagen, van der Hoeven et al. 2007, Einaudi, Gire et al. 2015). This implies that QoL should

not only include the well-being of the infant, but also the infants' future potential for independence. In practice, best interest assessments encompass medical considerations (objective components) and include moral values (subjective components) of the various stakeholders. For example, the moral status of premature infants is such a subjective component. Studies have shown that infants born extremely preterm were treated differently from term newborn infants, children and adults (Janvier, Bauer et al. 2007). But the majority of lay people would apply equal criteria for initiating intensive care treatment in preterm infants and children. This signals the need for further public and ethical discussion regarding the differential treatment of extreme preterm infants and resource allocation.

Often disagreement regarding the best approach to take is the result of different interpretations of what is best for the patient or results from prioritising interests differently (Rhodes and Holzman 2014). Parents' cultural or religious views can lead to persistent disagreement. Sometimes however, physicians are willing to withdraw or withhold life-sustaining treatment at the request of family's, despite their belief that continued treatment would be in the best interest of their patient (Moratti 2010). These complexities need to be further discussed and empirical research should explore whether a threshold of acceptable care could bypass the problems physicians are faced with in their daily clinical practice.

The diverging treatment choices found in our study may be caused by Switzerland's distinct cultural composition, as the French, German and Italian linguistic regions all have their own cultural and religious backgrounds. For example, liberal attitudes towards withholding intensive care were more evident among the French-speaking population. The German-speaking population was more likely to see the parents as the final decision-makers when there was a disagreement, while the French- and Italian-speaking populations felt that the physician should have more authority. Countries close to Switzerland, such as France, Germany and Italy, have also shown these cultural tendencies in neonatal healthcare providers or parents with regard to ethical decisions (Orfali 2004, Sauer, Dorscheidt et al. 2013, Gallagher, Martin et al. 2014). A Swiss study on survival outcomes of preterm infants also showed centre-to-centre differences, which were believed to be the result of a particular culture of NICUs (Steurer, Adams et al. 2015). Interestingly, both religion and personal experience with prematurity were less influential than anticipated in our study. They were only prominent in decisions to initiate intensive care treatment despite an expected low QoL. This suggests that the attitudes of the populations assessed in this survey might reflect the views and attitudes that would also be expressed by the parents of preterm infants in our

NICUs. As for religion, treatment decisions for neonates seem less related to religious values, as previously thought (Lantos 2015).

These socio-cultural differences are particularly important when they influence life and death decisions in very premature infants. In a context fraught with clinical and prognostic uncertainty, different moral interpretations are bound to happen. Despite the aim of most national guidelines to set clear boundaries for appropriate care, studies in several European countries have shown that treatment decisions reflect local NICU cultures (Gallagher, Martin et al. 2014). The process of coming to a moral consensus in a shared approach within pluralistic and diverse societies can be a difficult yet important one (Lantos 2016).

Some limitations of this survey should be considered. A general limitation of a telephone interview is that it does not allow for in-depth exploration and both the questions and responses are limited in their complexity. Despite the fact that the phone interviews needed to be relatively short, an open question was asked on the minimal criteria for an acceptable QoL. Furthermore, our respondents were categorised into one of the three language areas according to their area of residence. Finally, we did not ask people's specific religious denomination – such as Protestant, Catholic or Muslim – but focused on people's strength of belief in order to assess the potential influence of religion on attitudes and treatment decisions. Methodologically, our response rate was common for population-based telephone surveys on healthcare topics using quota sampling. Our findings can, therefore, be generalised to the whole nation. Notwithstanding the survey's limitations, our nationwide representative sample adds to the limited knowledge of the societal attitudes and values on treatment and EoLDM for extremely premature infants (Norup 1998).

In future research, it would be of interest to define a context where ethical trade-offs are accepted for decisions that cannot be considered medically beneficial or futile, but are somewhere in between. It is in this context where shared decision-making enables healthcare experts and parents to reflect on and reconcile with their underlying attitudes and values through continuous dialogue. Moreover, the dilemma of who should take the final decision when there is no consensus might disregard the underlying problem of assessing what is good care for extremely preterm infants. Meaningful support during EoL decisions requires optimal communication, which could limit the conflict and distress between the healthcare team and the family. The current understanding of morally pluralistic societies requires further discussion on the appropriateness of

advancing regulation in the field of highly specialised neonatal intensive care and on the soundness of ethical arguments that underlie EoL decisions.

Conclusion

Our survey reveals that the majority of the Swiss population clearly favoured shared decisions as the most appropriate decision-making model when faced with EoL decisions for infants of extremely low gestational age. Socio-cultural demographics, especially the linguistic region, influenced the attitudes toward extreme prematurity and decision-making. Our findings provide valuable information that can be used to understand the societal attitudes, values and arguments put forward by the parents of extremely preterm infants being cared for in the NICU. It also gives an insight into comparisons between, and within, countries that can fuel and stimulate debate in neonatal medicine, but also on a societal, public level.

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Conflicts of Interests

The authors have no conflicts of interest to disclose.

Exploring Societal Solidarity in the Context of Extreme Prematurity

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Author Contributions

All authors have contributed to the different parts of the manuscript, and each author has participated sufficiently in the work to take public responsibility for appropriate portions of the content. Miss Hendriks and Dr. Klein carried out the data extraction and analysis, interpreted the data, wrote the initial manuscript, and approved the final manuscript as submitted. Drs. Bucher and Fauchère designed the study, drafted the questionnaire, interpreted the data, critically reviewed and revised the manuscript, and approved the final manuscript as submitted. Dr. Streuli interpreted the data, wrote the initial manuscript, and approved the final manuscript as submitted. Dr. Baumann-Hölzle designed the study, interpreted the data, critically reviewed and revised the manuscript, and approved the final manuscript as submitted. All authors have read and approved the submission of the manuscript and take full responsibility.

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Abstract

Question: Extreme prematurity can result in long-term disabilities. It's impact on society is often not taken into account and deemed controversial. Our study examined the Swiss population's attitudes regarding extreme prematurity and people's perspectives regarding the question of solidarity for disabled people.

Methods: We conducted a nationwide representative anonymous telephone survey with 1210 Swiss residents, aged 18 or older. We asked how people estimate their own personal solidarity, the solidarity of their social environment and the solidarity across the country toward disabled persons. Spearman's correlation calculations were used to assess if a correlation exists between solidarity and setting financial limits to intensive care and between solidarity and withholding neonatal intensive care.

Results: For 36.0% of the respondents intensive medical care should not be withheld from extremely preterm infants, even if their chances for an acceptable quality of life were poor. For 28.8% intensive care should be withheld from these infants, while 26.9% held an intermediate position depending on the situation. 31.5% were against setting a financial limit to treatment of extremely preterm newborns with an uncertain future quality of life, 34.2% were in favour and 26.9% were deliberating. A majority (88.8%) considered their solidarity toward disabled people as substantial; the solidarity of their personal environment and of the society at large was estimated as high by 79.1% and 48.6%, respectively.

Conclusions: The Swiss population expressed a high level of solidarity, which may alleviate some pressure on parents and health care providers in the decision-making process in the neonatal intensive care units. In addition, there was no relationship between solidarity and people's willingness to pay for the care or withholding treatment of extremely preterm babies.

*Keywords***:** extreme prematurity; population survey; solidarity; disability; resource allocation; Switzerland

Introduction

Over the last decades, new treatment options have led to a decrease in mortality for extremely preterm infants. However, long-term morbidity has not yet decreased accordingly (Anderson, Baer et al. 2016, Holsti, Adamsson et al. 2016). It is often difficult to predict which infants will die, and which will survive with long-term disabilities. This medical and moral uncertainty complicates decision-making between parents and health care providers (Dupont-Thibodeau, Barrington et al. 2014). To this extent, the burdens for babies, families and health care professionals have frequently been studied, yet the impact on society is often not taken into account and deemed controversial. In fact, decisions do not only have developmental, psychological and emotional repercussions for extreme preterm infants and their families, but they also have an impact on society (Einaudi, Gire et al. 2015).

Providing care for infants at the limit of viability is one of the most expensive health care interventions (Korvenranta, Linna et al. 2010, Lantos and Meadow 2011). But in spite of these high costs, neonatal intensive care units (NICUs) are remarkably cost-effective (Caughey and Burchfield 2014, Partridge, Robertson et al. 2015). In addition, NICU survivors and their families often report a good quality of life (QoL) comparable to that of babies born at term (Roberts, Burnett et al. 2013, Huhtala, Korja et al. 2016). But discussions on resource allocation for infants born too early remain. Health care resources are limited and the question is how to fairly distribute limited resources amongst a patient population in light of long-term outcomes (Wilkinson 2013).

High individual costs, cost-effectiveness and resource allocation play a complex role in decision-making for extreme preterm infants. Besides immediate care parents may also consider society's acceptance and assistance much later in life when formerly extreme preterm infants with a moderate or severe disability may need continuous treatment and financial support (Sharman, Meert et al. 2005, Madrigal, Carroll et al. 2012). In this context, social cohesion as perceived by parents may influence choices about life-sustaining treatment. Societal solidarity, however, depends highly on the given circumstances of a country. Solidarity toward disabled people can identify the willingness to carry the costs for extremely preterm infants at risk for disabilities.

By means of a population survey we aimed to understand how people in different linguistic parts of Switzerland estimate their own personal solidarity, the solidarity of their social environment and the solidarity across the country toward disabled persons. We also intended to

analyse the relationship between solidarity and setting an upper limit to neonatal IC costs, and between solidarity and withholding IC for infants born extremely premature.

Methods

Study Design

We conducted a population-based telephone survey among Swiss residents on their opinions and values regarding extremely preterm infants. The questionnaire was prepared in German and translated into French and Italian. The translation accuracy was checked by back-translation into German and simultaneous review by a panel of translators to ensure identical semantic content in each language.

At the beginning of the interview, respondents were informed that our study concentrated on "extremely preterm infants", which meant infants born before 28 weeks of gestation – before 7 months of pregnancy with a birth weight often ranging between 600 and 800 grams. It was also clarified that these infants always require intensive care treatment.

The survey consisted of 16 questions with mean interview duration of 13.8 minutes (95% confidence interval 13.5–14.0 min). Themes that are explored in this article pertain to: (1) perceptions regarding extreme prematurity; (2) views on intensive care measures; and (3) assessment of the solidarity with disabled persons. Conventional sociodemographic information was collected on gender, age, residency, education and income level, as well as importance of religion, personal experience with prematurity and whether participants had children.

Data Collection

We recruited people living in Switzerland who were aged 18 years and older through random allocation drawn from the official telephone registry. Quotas were allocated for linguistic regions (German, French, and Italian), age and gender in order to allow for generalisation across the whole population. The market and social research institute gfs-zurich conducted the survey. Participants were informed about the study and procedures for anonymity. Respondents had the opportunity to withdraw from the survey at any time point. As this anonymous population survey did not include any patient data or health-related data of the participating persons, the Ethics Committee of the Canton Zurich did not require formal ethical approval.

Data Analyses

Statistical analysis was performed using IBM SPSS Statistics 22 (Armonk, NY, USA). The quota from the Italian-speaking part was increased in order to have a sufficient number of answers for comparisons with the other linguistic areas. All cases were weighted to adjust for this stratification. Thus, the percentages presented cannot be derived from the absolute unweighted numbers in the tables. Groups (age, gender, linguistic region, importance of religion, level of education) were compared using a z-test or a t-test with Bonferroni adjustment for multiple comparisons where appropriate. Linear regression analysis to test for the independent effect of these factors was considered but discarded, since this would have meant use of weighted data, in which case the proportion from the Italian-speaking part would have been too small to detect any differences. A p-value <0.05 was considered statistically significant.

Solidarity was assessed with a 5-point Likert scale. These responses were grouped into two adjacent categories in the text for ease of interpretation: high (somewhat or much higher estimation) or not high (average or lower estimation). Spearman's correlation calculations were used to assess if a correlation exists between solidarity and setting financial limits to intensive care, and between solidarity and withholding intensive care treatments.

Results

Characteristics of the Respondents

A total of 1210 adults living in Switzerland completed the telephone survey, yielding a 23.7% response rate. Most respondents were aged between 40 and 64 years (45%) and had children (70%). Two thirds (68%) did not have previous experience with prematurity personally or in their social surrounding (extended family and/or friends). Five per cent of respondents were parents of a premature infant and 26% knew parents of a preterm infant in their close social environment. Religious faith was reported to be stronger in the German (54.1%) and Italian regions (53.3%) as opposed to the French region (41.6%). The main characteristics of our respondents are summarised in Table 1.

Table 1. Characteristics of respondents in the survey

Characteristic	%	N
Gender		
Male	49.7	602
Female	50.3	608
Region[a]		
German part	72.0	707
French part	24.0	303
Italian part	4.0	200
Age		
≤ 39 years	35.0	423
40-64 years	45.0	544
≥ 65 years	20.1	243
Having own children		
Yes	70.2	837
No	29.8	373
Personal experience with prematurity		
Weak	68.2	831
Strong	31.5	375
Missing	-	4
Importance of religion		
Not important	48.9	586
Important	51.1	606
Missing	-	18

Characteristic	%	N
Education		
Low	7.2	99
Middle	66.1	805
High	26.6	297
Missing	-	9
Monthly income (CHF)		
≤ 4000	13.8	176
4001 - 7000	34.2	399
7001 – 11000	25.9	308
> 11000	11.9	134
Missing	-	193

Total N=1210. Percentage without missing values unless indicated.

[a] Regions are based on: the German speaking part, the French speaking part and the Italian speaking part of Switzerland. All results in % are weighted for regions, numbers of respondents (N) are unweighted.

Extreme Prematurity and Intensive Care

In general, people's associations with extreme prematurity were very diverse. The most common impressions were linked to notions such as fighting for survival (11%), future consequences (9%), incubators (9%), giving them a chance (7%) and small infants (7%). Fewer answers addressed emotions (≤4%: pity, hope, fear, burden) or the QoL and disabilities (≤3%). The majority of our respondents (67.1%) estimated that the use of highly specialised medicine for these extremely preterm infants would be supported in Switzerland. A smaller number (20.8%) thought intensive care would not be endorsed, and 12.1% did not know.

For 36.0%, intensive medical care should not be withheld from extremely preterm infants, even if their chances for an acceptable QoL were poor. For the same situation, 28.8% thought intensive care should be withheld from these infants, 26.9% held an intermediate position depending on the individual situation, and 8.3% did not know. Regional differences were found. More respondents from the French-speaking part of Switzerland (39.3%) agreed to withhold

treatment as opposed to the German-speaking (25.9%) and the Italian-speaking (18.5%) regions ($p < 0.001$).

An upper financial limit for treating extremely preterm infants with an uncertain future QoL was considered justified by 34.2% of respondents. Another third (31.5%) considered such a financial limit as unacceptable, another 26.9% took an intermediate view, and 7.4% could not answer the question. Higher educated people (42.9%) were more often against an upper financial limit than people with a medium (28.0%, $p = 0.003$) or lower (25.3%, $p < 0.001$) level education. People who considered that intensive care treatment should be withheld from extremely preterm infants with an expected poor QoL more often agreed to set financial limits to intensive care, as shown by a weak correlation ($r = 0.125$, $p < 0.001$; Table 2).

Table 2. Correlations between withholding treatment, setting an upper financial limit, and solidarity

		Medical interventions should be withheld when prospective outcome is poor	An upper financial limit for intensive care is justifiable	My solidarity towards disabled people is substantial.	The solidarity towards disable people in my personal environment is substantial.	The solidarity towards disabled people in Switzerland is substantial.
Medical interventions should be withheld when prospective outcome is poor	r		0.125	-0.054	-0.060	0.016
	p		**<0.001**	0.104	0.070	0.627
	N		1042	1107	1082	1082
An upper financial limit for intensive care is justifiable	r	0.125		0.000	-0.020	0.112
	p	**<0.001**		0.991	0.551	**0.001**
	N	1042		1114	1087	1095

		Medical interventions should be withheld when prospective outcome is poor	An upper financial limit for intensive care is justifiable	My solidarity towards disabled people is substantial.	The solidarity towards disable people in my personal environment is substantial.	The solidarity towards disabled people in Switzerland is substantial.
My solidarity towards disabled people is substantial.	r	-0.054	0.000		0.517	0.162
	p	0.104	0.991		**<0.001**	**<0.001**
	N	1107	1114		1171	1174
The solidarity towards disable people in my personal environment is substantial.	r	-0.060	-0.020	0.517		0.322
	p	0.070	0.551	**<0.001**		**<0.001**
	N	1082	1087	1171		1149

		Medical interventions should be withheld when prospective outcome is poor	An upper financial limit for intensive care is justifiable	My solidarity towards disabled people is substantial.	The solidarity towards disable people in my personal environment is substantial.	The solidarity towards disabled people in Switzerland is substantial.
The solidarity towards disabled people in Switzerland is substantial.	r	0.016	0.112	0.162	0.322	
	p	0.627	**0.001**	**<0.001**	**<0.001**	
	N	1082	1095	1174	1149	

Spearman's correlation coefficients are shown. Results are weighted for regions, numbers of respondents (N) are unweighted. For exact questions and answer categories, see supporting information.

Table 3. Evaluations of solidarity

Do you agree with the following statements?	1 completely disagree		2		3 agree partially		4		5 completely agree		do not know	
	%	N	%	N	%	N	%	N	%	N	%	N
My solidarity towards disabled people is substantial.	0.7	7	1.1	13	8.7	103	32.5	404	56.4	676	0.6	7
The solidarity towards disabled people in my personal environment is substantial.	1.2	14	2.3	28	14.5	187	33.5	415	45.6	530	2.9	36
The solidarity towards disabled people in Switzerland is substantial.	2.6	32	9.5	104	37.1	427	28.8	360	19.7	256	2.2	31

Total N=1210. All results in % are weighted for regions, numbers of respondents (N) are unweighted.

Self-Rated and Estimated Solidarity with Disabled People

The population's perception toward solidarity for people with disabilities was explored. We asked respondents to estimate their own solidarity, the solidarity in their personal environment (i.e., family and friends), and finally the solidarity within the Swiss population. Overall, 88.9% of respondents expressed substantial solidarity towards disabled people. People also considered the solidarity of their social environment as high (79.1% of respondents). Solidarity towards disabled people in Switzerland was felt to be substantial by 48.5% of our sample (Table 3). The estimation of one's own personal solidarity was influenced by several sociodemographic factors. Respondents with a strong affiliation to religious faith more often considered their solidarity with disability as substantial (63.1%), in contrast to people without a religious affiliation (49.3%, $p<0.001$). Likewise, parents (62.7%) rated their personal solidarity higher than non-parents (41.6%, $p<0.001$). The same held true for people residing in the German-speaking region (60.7%) who rated their personal solidarity higher than people from the French-speaking area (45.6%, $p<0.001$). For 58.5% of the respondents the willingness of society to support disabled people had improved over the last 10 years. A smaller group (25.7%) held the view that the support has remained constant, whereas 10.2% considered it has deteriorated.

Spearman's correlation analyses revealed no correlation between people's self-rated solidarity towards disabled persons and rejecting the possibility to withhold intensive care treatment in cases with poor prognosis (Table 2). In addition, no correlation was found between respondents' estimations of their personal solidarity and setting financial limits for the treatment of extremely preterm infants. Similarly, solidarity of the respondents' social environment also did not show any of the above-mentioned correlations. People who found an upper financial limit for treatment justified, rated solidarity toward disabled persons in Switzerland as more important, although this correlation was weak ($r = 0.112$, $p = 0.001$).

Discussion

This is the first nationwide study in Switzerland and the largest representative report to analyse public perceptions and values toward extreme prematurity (Goldnagl, Freidl et al. 2014). Our major finding is that the Swiss population expressed a high overall solidarity toward disabled persons. Interestingly, there was a clear difference between the personal, social environmental and societal level of solidarity. Swiss people rated their personal solidarity as very high, whereas they estimated

that solidarity in their social surrounding and in the general society was perceived as lower. This could be caused by the fact that people are better at estimating their own valuations as opposed to those of the society. Conversely, response and social desirability bias could also have caused these differences. Furthermore, we found that people who considered it justifiable to withdraw care for babies born extremely preterm with expected poor QoL outcomes were also more inclined to set economical limits to IC for this group of patients. However, those people who found financial limits justified estimated that the solidarity in Switzerland was high.

We can only speculate on the reasons for such a high solidarity. On the one hand, this might portray a commitment to a fair distribution of health care resources. Despite the need for resource allocation and setting financial limits, it is possible to do so fairly while still protecting the health interests and needs of the weakest members in the society. On the other hand, it might illustrate the limited role financial and economic factors play in people's self-rated solidarity toward weaker groups in society. The motivation of individuals to support health care and social protection has mainly focused on financial aspects, however, these considerations are maybe also influenced by elements of compassion, commonality and mutuality (Ter Meulen and Houtepen 2012). This can go beyond economic considerations (Prainsack and Buyx 2012). Instead it might be the common ground between individuals from which mutual obligations arise to help and support each other when necessary (Bayertz 1999). Hence, our study indicates that solidarity does not merely reflect the commitment of a group to carry the financial costs.

Another reason that might explain the high level of solidarity people emphasised is the resemblance of the vulnerability of disabled patients such as former extreme preterm infants. It is known that solidarity is strongest with those people or groups that strike us as noteworthy and with whom we share resemblance (Rorty 1989, Bayertz 1999). The extreme vulnerability of extreme preterm infants and their acute need for life-sustaining care could foster a connection of resemblance (Cadore, Boitte et al. 2000). Additionally, these infants cannot be held responsible for the medical decisions that have taken place either prior to their birth (i.e. decisions to initiate/withhold treatment) or after birth (e.g. continuing/withdrawing life-sustaining therapy).

Finally, the context of the Swiss health care system could have supported the right conditions for a high level of solidarity. The Swiss health care system is thought to reflect an egalitarian sentiment where solidarity plays an important role in public discourse. In addition, the health system and the current social health insurance system are greatly valued by patients and the population as a whole (Biller-Andorno and Zeltner 2015, De Pietro and Crivelli 2015). Hence,

solidarity is something that is shaped within a society over time and can vary in different contexts (Rorty 1989). This is also apparent in other countries where resource allocation decisions are based on medical facts as well as societal values (National Institute for Health and Care Excellence 2014).

The geographic variation within Switzerland raises the question as to whether there is an explanation based on factors such as religion, age, sex and socioeconomic status. Age and gender were evenly distributed among the linguistic regions as our survey was designed to be representative for these variables. The importance of religion varied between the linguistic areas, being strongest in the German- and Italian-speaking regions. Income and education were correlated and were higher in the German part. However, our analyses do not indicate that the geographic variation would be due to the differences observed in importance of religion or socioeconomic status.

Our findings potentially impact on parents' decision-making in the context of extreme prematurity. One study on parental decision-making preferences has suggested that the level of societal support and financial difficulties could be aspects that influence parents' decision-making (Madrigal, Carroll et al. 2012). Another study investigating what would influence parents' decisions to limit or withdraw life support has pointed out that personal finances and societal limitations in health care resources are a concern for parents (Sharman, Meert et al. 2005). Despite national policies specifying that economic considerations should not interfere with ethical decision-making in individual cases (Nuffield Council on Bioethics 2006, Berger, Bernet et al. 2011), clinical reality shows that sometimes issues unrelated to the child's well-being – such as social acceptance and support – may influence decision-making (Sharman, Meert et al. 2005, Cornfield and Kahn 2012, Madrigal, Carroll et al. 2012). Under these circumstances, parents together with the health care team must make decisions about the appropriateness of life-prolonging IC treatment. However, assessments of societal support and solidarity will become more important with recent calls for a sustainable health system.(Biller-Andorno and Zeltner 2015) Resource allocation has many inherent ethical concerns that are beyond the scope of this paper (Savulescu 2002, Camosy 2011, Wilkinson and Savulescu 2014), but, of relevance to this discussion, the more a health care system redistributes resources to the members of the collective, the more it is suggested to be an articulation of solidarity (Prainsack and Buyx 2012, Ter Meulen and Houtepen 2012).

Strengths and Limitations

Our study has several limitations. A general limitation of a telephone interview is that it does not allow in-depth exploration, and both questions and responses are limited in their complexity. Despite the required time shortness of phone interviews, several open questions were asked on perspectives concerning extremely preterm infants and minimal criteria for an acceptable QoL. Furthermore, our respondents were categorized in one of the three language areas according to their current area of residence. This begs the question whether some respondents were born and raised or simply moved into their current area of residence. A further limitation of our study includes the complexity of the notion of solidarity. Due to the time constraints of a telephone survey, solidarity could not be clearly defined to respondents prior to the survey. Therefore, it is most likely that respondents may have understood only the most general notion of the term.

Limitations notwithstanding, our nationwide representative sample extends a limited knowledge of the societal solidarity for infants born extremely premature. Methodologically, our response rate is common for a telephone survey of the population on health care topics using quota sampling. Our study sample is representative of the demographic distribution of Switzerland as portrayed by the Federal Statistics Office. Therefore, our findings can be generalized to the population, thereby allowing a conclusion on how the Swiss population perceives extreme prematurity, and on how end-of-life decisions should be taken for this patient group. Future work should further analyse the validity of solidarity and take into account the limitations encountered in our work. Also, the relationship between solidarity and economic factors should be extended and should include an investigation of the link with social and emotional acceptance and support (Prainsack and Buyx 2012).

Conclusion

Our survey evaluated the societal attitudes toward people living with disabilities in the context of extreme prematurity. Caring for a disabled person always implies the use of health care resources and is directly linked to societal solidarity and support. According to our study, the populations' high level of solidarity toward disabled people goes beyond the financial focus often taken when assessing solidarity. This knowledge is of paramount importance in the ongoing societal discussion on allocation of heath care resources. Even more importantly, a high societal cohesion toward disabled persons may alleviate some pressure on parents in the decision-making process regarding their extremely premature infant in the NICU.

Conflicts of interest

The authors have no conflicts of interest to disclose.

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PART II PARENTAL EXPERIENCES

Parents' Bonding with their Extremely Preterm and Dying Child

Andrea Abraham* [1,2] and Manya J. Hendriks* [1,3] on behalf of the NRP67 Neonatal End-of-Life Study Group

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Author Contributions

All authors contributed to the different parts of the study. All authors designed the study and drafted the interview guide. Mrs. Hendriks and Dr. Abraham carried out the data collection and analysis. Both authors interpreted the data and contributed to the initial manuscript. All authors revised and approved the final manuscript. All authors have participated sufficiently in the work to take public responsibility for appropriate portions of the content.

Additional members of the NRP67 Neonatal End-of-Life Study Group include the following: Jean-Claude Fauchère, Hans-Ulrich Bucher, Ruth Baumann-Hölzle, and Thomas Berger.

Andrea Abraham & Manya J. Hendriks, on behalf of the Swiss Neonatal End-of-Life Study Group (2017), "You Can Only Give Warmth to Your Baby When It's Too Late": Parents' Bonding with their Extremely Preterm and Dying Child. *Qualitative Health Research,* n/a:n/a. doi:10.1177/1049732317721476

Abstract

This study on end-of-life decisions in extremely preterm babies shows that the parents under study experience a multitude of stressors due to the immediate separation after birth, the alienating setting of the neonatal intensive care unit (NICU), the physical distance to the child, medical uncertainties, and upcoming decisions. Even though they are considered to be parents (assigned parenthood), they cannot act as primary caregivers. Instead, they depend on professional instructions for access and care. Embodied parenthood can be experienced only at the end-of-life, that is, during the dying trajectory and after the child's death. Professionally supporting parents during this compressed process (from assigned and distant to embodied parenthood) contributes fundamentally to their perception of being a family and supports their mourning. This calls for the further establishment of palliative and bereavement care concepts in neonatology.

Keywords: extreme prematurity, neonatal intensive care unit, end-of-life, embodied parenthood, parental bonding, palliative and bereavement care, qualitative content analysis, Switzerland.

Introduction

In Switzerland, 0.3% to 0.4% of all babies are born extremely preterm, meaning that they are born between 22 and 27 weeks of gestation.[1] Due to medical progresses, survival rates of infants born at the limit of viability have increased over the last decades. Berger, Steurer et al. (2012) reported an average survival rate in Switzerland (2005–2009) ranging from 4% at 23 weeks to 88% at 27 weeks of gestation. A very recent study has assessed the reasons and circumstances of deaths in this population between 2012 and 2015 (Berger, Steurer et al. 2017). Results show that out of 594 deaths, 280 (47%) were stillborn, 185 (31%) died in the delivery room (DR), and 129 (22%) died in the neonatal intensive care unit (NICU). The principal causes of death in the NICU were severe congenital malformations, sepsis, cardiovascular failure, respiratory failure, gastrointestinal complication, and neurologic complications. Yet extreme prematurity represents not only a clinical challenge but also a complex and stressful experience for the parents concerned (Aagaard and Hall 2008, Cricco-Lizza 2014, Petteys, Goebel et al. 2015). Even though they are considered to be parents, their parental "role begins and evolves in a public and medically focused context" (Flacking, Ewald et al. 2006, p. 71) in an "over-populated hospital space" (Hall and Brinchmann 2009, p. 131). Parental bonding (i.e., physical, social, and emotional relationship-building interactions between parents and baby) is especially difficult when babies do not survive and die either in the DR or in the NICU. This process is strongly compressed and complicated by several factors.

First, studies show that parents who experience the birth of an extremely preterm baby do not feel mentally prepared for this situation (Hall, Kronborg et al. 2013). They enter a world foreign to them, encountering the NICU, with its sounds, incubators, wires, and tubes, as a highly technologized environment (Lundqvist, Nilstun et al. 2002, Shin and White-Traut 2007, Cleveland 2008, Hutchinson, Spillett et al. 2012, Shattnawi 2015). This open space immediately "signal[s] life and death" (Flacking, Ewald et al. 2006, p. 73). Parents enter a "bubble," which "only a few outside this bubble [are] able to grasp and understand" (Flacking, Ewald et al. 2006). Faced with this unfamiliar setting, parents have reported highly diverse emotional reactions (Owens 2001, Orfali and Gordon 2004). These range from the uncertainty of the baby's survival, possible medical complications and future disabilities, to feelings of non-familiarity, confusion, disappointment, distress, shame, failure, guilt, and helplessness (Schappin, Wijnroks et al. 2013, Fortney and

Steward 2015, Hoffenkamp, Braeken et al. 2015). The situation in which they find themselves does not correspond at all with their expectations of parenthood (Shin and White-Traut 2007).

Second, even though parents are more involved in caring for their preterm baby nowadays, the degree of this involvement remains contested in some NICUs (Nyqvist and Engvall 2009). Parents' active participation actually depends extensively on the NICU culture and on the attitudes of health care professionals (HCPs; Fenwick, Barclay et al. 2001). Some studies illustrate lacking family-centred care and parents' missing or limited involvement (Browne 2003). For instance, mothers in Shin and White-Traut (2007) study experienced "disconnection, detachment, disaffection and disengagement" (p. 93). In these cases, HCPs have remained the primary caretaker (Fenwick, Barclay et al. 2001). One effect described by parents is that they "felt as though they were parenting from a distance and struggled with feelings of uselessness while yearning to be useful in baby care" (Cleveland 2008, p. 672). Linked to this, parent–baby bonding in the first NICU weeks often strongly depends on NICU culture and on nurses' support and does not simply occur naturally (Shin and White-Traut 2007, Fenwick, Barclay et al. 2008, Guillaume, Michelin et al. 2013, Currie, Christian et al. 2016). Rather, it requires special awareness among NICU staff to "facilitate the parents' independence as caretakers" (Eden and Callister 2010, p. 37).

A third factor complicating bonding is limited physical contact with the baby. Holding or touching the child has been described as a turning point in parents' NICU experience (Shin and White-Traut 2007, Cleveland 2008, Hutchinson, Spillett et al. 2012, Guillaume, Michelin et al. 2013). Guillaume et al. (2013) mentioned that physical closeness and "increased knowledge with contact" (p. 4) are crucial in helping parents to bond with their baby, a finding confirmed by other studies (Cleveland 2008). However, it requires professional instructions to ensure the baby does not get hurt (Shin and White-Traut 2007, Guillaume, Michelin et al. 2013, Shattnawi 2015). When physical contact is barely possible, child–parent interaction is relatively nonreciprocal. Shattnawi (2015) revealed that this physical distance is extremely stressful for mothers. Flacking et al. (2006) illustrated how mothers felt "they had become mothers physically but not socially or emotionally" (p. 73). This detachment leads to parents describing themselves as passive (Guillaume et al., 2013), as "watchers" (Flacking et al., 2006, p. 75), as "just a visitor" (Flacking et al., 2006, p. 74), or as "strangers" (Shattnawi, 2015, p. 79).

These factors mirror parents' potential struggle to find their role and space in the NICU (Flacking et al., 2006) and to actually make sense of their new role. Shattnawi (2015) and Watson

(2011) conceptualized the transition to parenthood within the NICU environment as a liminal phase. Liminality, as a social scientific concept, implies a transitional in-between process (Rapport and Overing 2000). Parents reporting their experience of having a baby in the NICU often describe the process of "bonding," which gradually transforms their initial feelings of alienation or unconnectedness with their baby into the more typical emotions associated with full-term babies and with "feeling like a parent" (Shin and White-Traut 2007, Cleveland 2008, Forman 2009, Hutchinson, Spillett et al. 2012, Guillaume, Michelin et al. 2013, Fei 2015).

This article illustrates the perspective of parents whose extremely premature baby died within a few hours or days after birth. It focuses on how parents experience the dying trajectory of their extremely premature babies in the NICU, how they experience parenthood, and how HCPs can facilitate parent–child bonding in the very short time available. It presents data from a qualitative study on end-of-life decision making in extremely preterm infants at the limit of viability. Our study was secondary to a larger quantitative survey on end-of-life decisions in extremely preterm infants. Our task was to shed light on the parental perspective, whereas the other studies forming part of the survey focused on the perspectives of society, of NICU HCPs, and on ethical questions arising from the results (Hendriks, Bucher et al. 2017, Hendriks, Klein et al. 2017, Klein, Bucher et al. 2017).

Methods

This study used a methodological approach based on symbolic interactionism with its focus on the reconstruction of subjective meaning (Flick 2014). For data collection, we chose an interview design with ethnographic aspects (temporary field stays, minimal observations). We conducted a retrospective study (and not, for instance, a prospective ethnographic study) due to the assumed mourning process of parents. During our informal talks in the preparatory field stays, we were advised not to conduct interviews with parents who had only recently lost their child (Rosenblatt 1995, Hellmann, Williams et al. 2013). The intimate and sensitive character of the mourning process also kept us from doing participant observation when parents were present in the confined NICU spaces. Due to this "dilemma of discretion" (Anspach and Mizrachi 2006, p. 63), our main data consist of interviews. We are aware of the fact that these methodological decisions produced data potentially affected by our distant perspective on NICU events and by parents'

blurred memories. Concerning the latter, it proved beneficial to interview couples because they co-constructed their experience.

Participants

We applied the technique of criterion-based, purposive sampling. Forty-two potential participants (parents who lost an extremely preterm, live-born child in the years 2013–2015 at the tertiary-level NICU of an urban, university teaching hospital) received a letter from their former neonatologists soliciting their participation in the study. Written in two languages (German and English), the letter explicitly invited parents from different cultural backgrounds to participate. Due to ethical considerations, we refrained from calling people up and sent a reminder instead. We then interviewed those 20 parents (seven couples, one father, and five mothers) who contacted us (see Table 1 for demographics).

Table 3. Infant Characteristics

Characteristics		**N**
Infants Sex	Male	7
	Female	6
Infants Age	22 0/7-6/7	1
	23 0/7-6/7	1
	24 0/7-6/7	5
	25 0/7-6/7	4
	26 0/7-6/7	-
	27 0/7-6/7	2
Weight in Grams	250-500	3
	500-750	6
	750-1000	2
	>1000	1

Characteristics		N
Number of Survived Hours/ Days	Hours	6
	<5 days	2
	5-10 days	3
	10-15 days	2
Cause of Death	Extreme Prematurity	5
	Intra-uterine growth restriction	1
	Respiratory Disease	3
	Necrotizing Enterocolitis	1
	Major IVH	3
Place of Death	Delivery Room	6
	NICU	7

Informed Consent

Before the interviews, parents received written and oral information about the study. They were ensured anonymity and provided informed consent. During the interviews, we also conducted "process consenting" (Hutchinson, Spillett et al. 2012), that is, regularly establishing participants' willingness to proceed with the interview, whether they needed a break, or whether they preferred to terminate it. Even though most parents were very emotional, none decided to terminate the interview earlier than planned and expressed their gratitude for having the opportunity to tell their baby's story (Rosenblatt 1995, Orfali and Gordon 2004, Buckle, Dwyer et al. 2010).

Data Collection

We placed great emphasis on detailed preparation, spending time in the NICU, talking to staff members (e.g., neonatologist, physiotherapist, psychologist, pastor, music therapist), and meeting researchers experienced in this particularly sensitive area. We also spent some time in virtual spaces (e.g., chat forums) used by affected parents. However, these efforts did not generate data

that we analysed further but rather contextual information that we used to enhance the sensitivity and adequacy of our data collection method (Currie, Christian et al. 2016). We conducted a total of 13 in-depth interviews at a place chosen by the 20 parents. Both researchers were present at all interviews. Five interviews were conducted at home, three at the university hospital, four at Dialog Ethics, and one at the mother's place of work. We initiated the interviews by appreciating parents' willingness to share their experience and invited them to tell their baby's entire story in chronological order with a particular focus on end-of-life decisions. Some parents also showed us photographs, diaries, and other memories of their children. After the in-depth interviews, we asked parents to clarify specific topics and added a semi structured interview if we felt that certain topics had remained unaddressed. The interviews were tape-recorded and lasted between 60 and 160 minutes (97 minutes on average).

Data Analysis

Data analysis followed the principles of hermeneutically oriented qualitative content analysis (Kuckartz 2014). On one hand, the limited time available required an efficient, team-oriented, and well-structured working procedure. On the other, we had to generate descriptive results enabling comparisons with the two preceding quantitative studies. However, we also wanted to conceptually elaborate on topics emerging during the research process (e.g., parenthood). We therefore chose a methodological approach allowing for descriptive and interpretative analysis. Kuckartz's approach allowed us to also work with theoretical memos and to develop conceptual models on identified key topics. It roughly includes the following steps: inductive-deductive generation of thematic categories, first rough coding, further development and differentiation of categories, coding of all transcripts, category-based analysis, and writing of the research report.

Completed interviews were transcribed verbatim. First, four transcripts were coded by both authors to achieve congruent coding practice using the qualitative data analysis software MAXQDA. Second, transcripts were randomly divided between the two researchers and coded along a coding scheme comprising a chronological axis (pregnancy, birth, liminal phase, end-of-life, dying, and death) and a thematic axis (e.g., experience, values and attitudes, support, communication). The codes were developed deductively (based on our research questions) and inductively (topics emerging from preparations and interviews). Third, the codes were summarized in a descriptive manner, whereas interpretative notes were formulated as theoretical memos. In the course of coding, clustering codes into categories, and writing the theoretical memos, we moved

from merely describing to generating a conceptual model of parenthood (Bendassolli 2013, Kuckartz 2014, Silverman 2015).

All codes, summaries, memos, and running texts were cross-checked and complemented by the other researcher. Discrepancies were resolved through repeated discussion. Finally, the codes were systematized and categorized (Silverman 2015). Running text was written for each category. Besides closely collaborating on each methodical step, we searched for additional rigorous procedures. We continuously compared our results with those of the larger project's primary studies, even though the different data sets were not systematically triangulated. In addition, following data collection, we conducted five interviews with HCPs from the NICU under study (two neonatologists, one obstetrician, one NICU nurse, and one midwife). We found it important to reflect on the collected data with the help of HCPs experienced with working with parents in such situations. As we did a retrospective study and did not experience any dying situations during our NICU stays, we needed (a) some kind of validating feedback to our ongoing interpretation of the collected data, (b) answers to questions that we did not dare pursue in-depth with parents, and (c) information on "backstage"-procedures invisible or unknown to parents.

We reached thematic saturation within and across interviews for a very particular type of parents (extremely preterm, live-born infants who survived only for a short time). This means that we obtained no new information and could not further enlarge our coding scheme and categories (Bowen 2008, O'reilly and Parker 2013, Fusch and Ness 2015). Even though the richness and thickness of our data outweighed the small sample size, our explanatory model is hypothetical in nature and needs to be critically assessed in further studies, for instance, involving prospective ethnographic designs or parents from diverse cultural backgrounds (Brooks, Holdtich-Davis et al. 2016). As mentioned, our sampling strategy was unable to reach such parents.

Ethical Considerations

The Ethics Committee of the Canton of Zürich assigned the study a declaration of no objection (case number 64-2015). Even though we obtained this declaration, we felt a strong ethical responsibility to prepare and conduct our study as carefully as possible (Stroebe, Stroebe et al. 2003). We paid particular attention to recruitment (form, formulations, timing and reminder), the pain our interview request and the meeting might cause, verbal and nonverbal communication during the interview, emotional limits during the interview, processual consenting, the slippery

transition between interview and therapy, and our own emotional involvement before, during, and after interviews (Rosenblatt 1995, Stroebe, Stroebe et al. 2003).

Results

The extremely premature babies of the interviewed parents were born alive at 22 to 27 weeks of gestation (M_{age} = 24 6/7 weeks of gestation) and with a birth weight ranging from 340 to 1100 g (M_{weight} = 640 g). We distinguished two dying trajectories (Figure 1): First, those live-born children who died in the DR right after birth. For these children, intensive care was withheld and replaced by comfort care. Intensive care was not initiated either because the child was below the hospital's defined limits of initiation of neonatal intensive care or because of a late-term abortion. Second, those infants who received intensive care and who were then transferred to the NICU where they died within a very short time after intensive care was withdrawn due to medical futility. The latter group will be subject of the following results. Results concern parenthood in two phases of the dying trajectory: (a) after the baby's transfer to the NICU (a phase of uncertainty) and (b) the end-of-life phase (when death is certain).

Phase of Uncertainty

The first phase after the baby's admission to the NICU is characterized by uncertainty regarding the further clinical course and by parental bonding being hampered by numerous factors. This section discusses how parents cope with the limited options for bonding with their baby.

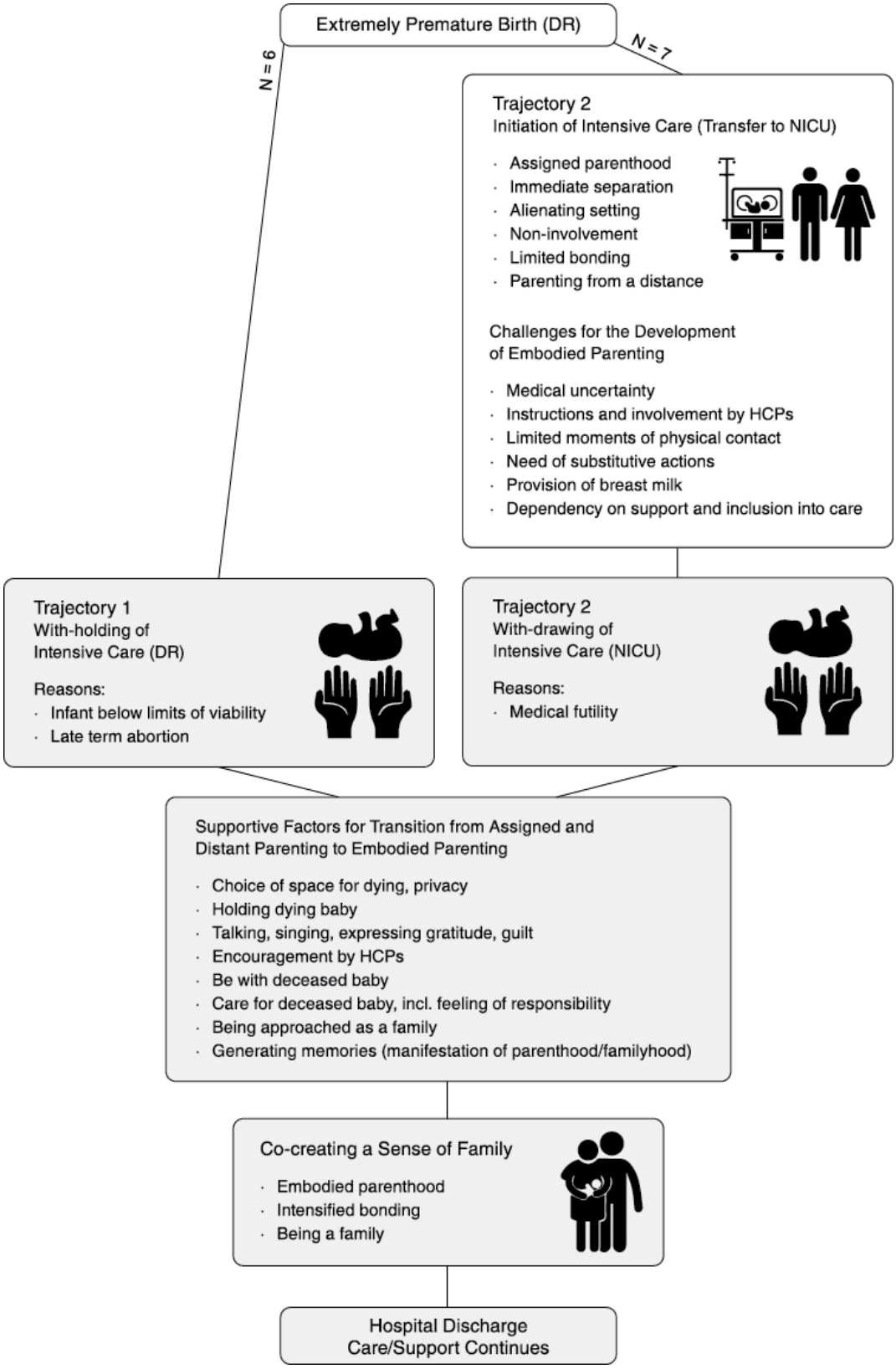

Figure 1. Conceptual model illustrating the process from assigned to embodied parenthood at the end-of-life. *Note:* DR = delivery room; NICU = neonatal intensive care unit; HCP = health care professional.

Immediate Separation of Parents and Baby after Birth

After mothers went through (a possibly traumatic) delivery, they were separated immediately from their child. Sometimes, however, the father was able to have a look at the baby before or during the NICU team's examinations. After these first intensive care interventions, the baby was transferred to the NICU. All parents whose baby was transferred to the NICU reported that they were strongly encouraged by the HCPs to visit their baby as quickly as possible after birth. Mothers who underwent a caesarean or who suffered a different birth-related burden were even brought to the NICU in their beds:

> These deeply human moments were genuinely fostered. I asked: "To the NICU in bed? This can't be possible." But they [HCPs] said: "You must see your baby" and took me there in my bed. (Mother)

The fathers would act as go-betweens or messengers between baby (NICU) and mother (maternity ward) if the mother was not yet mobile. If mothers were not allowed to visit their baby at the NICU (e.g., due to an intervention), or if HCPs did not have time to take them to their baby, this lack of access was perceived as very disturbing.

The separation of mother and child involved two different care settings: the maternity ward and the NICU. Mothers reported how challenging they experienced the NICU's one-sided focus on their baby's well-being:

> Of course the NICU only covered the child's part; the mother's or the parents' part doesn't really exist. You find yourself in an air bubble. Depending on how you manage to compensate this mentally, you can have quite a problem there. (Mother)

Limited access to the baby was intensified whenever the mother left hospital while the baby remained in the NICU. Parents experienced traveling back and forth to the NICU as an additional stressor.

Entering the Alienating NICU Setting

From delivery on, parents found themselves in a dissociated state of mind, which they described as a "cloud," "film," "dream," and/or "shock." Parents struggled to overcome their feelings of detachment, disconnection, and distance. They felt as if they were not really "there." They experienced mixed feelings: joy and love on one hand, uncertainty, sadness, guilt, and

disbelief on the other. Experiencing this emotional rollercoaster led to different attitudes, values, and expectations among parents. For instance, one couple found it inappropriate that the HCPs congratulated them on their baby because they "did not know what would happen" (Father). In addition to the unexpectedness of preterm birth and the feeling of being unprepared, parents perceived entering the NICU setting as alienating. The highly technological environment (incubators, computers, cables and tubes, the medical-technical interventions on the child) and the multitude of different people moving within the NICU (HCPs, other staff, other families) heightened this perception. Parents needed instructions on how to enter and move about in this unfamiliar setting. Their sense of alienation was further enhanced by the baby's visual appearance (size and frailty of the body, red skin colour, oxygen mask, and other apparatuses attached to the baby). They had to get used to this unusual appearance.

Visiting the baby in this environment was sometimes experienced as a burden. Some mothers reported that at times they did not have the energy to visit their baby.

> I once saw a TV interview with a mother whose experience was exactly the same as mine: She said that the maternal feelings came right away, despite everything, despite the incubator, which is a barrier. You cannot hold your baby, you can only touch her a little bit, but with sanitized hands. This is totally different than how you envisaged motherhood and these feelings . . . There's love and sadness, all together. It's true that it's your daughter right away. But it's horrible to see this baby so wired. I don't know where there weren't any wires. With this mask, with this tube to support the lungs. And to see her skin, which was so thin and constantly needed to be oiled and cared for. To see how she was still almost translucent. All these little details showed how frail she was. I admit that I even had moments when I visited her but had to leave. I was doing so badly. I couldn't look at her like this. And then I found courage again and said "No, she's my daughter, I'll go back to her again." (Mother)

However, parents also described how in some cases they perceived implicit expectations of the NICU staff about the frequency of their visits despite their frail physical or mental state. For

instance, one mother remembered that when her child was dying a nurse said that it was better that the baby was dying as the mother would not have had the strength to get through the NICU period.

Limited Physical Experience of the Baby

Despite parents' access to the NICU, the incubator and the technological setting represented a fundamental physical barrier between the baby and its parents and intensified parents' perception of the situation as unreal. Parents had difficulty realizing that the baby in the incubator was their own child. The physical barrier caused suffering because parents could not hold their child but only touch it gently due to its frailty or the risk of infections. Retrospectively, parents highly appreciated those moments in which direct physical contact was possible.

> It's one of my nicest memories that I was reaching [into the incubator] and the little one was holding my finger with his little hand. That was beautiful. (Mother)

One mother, however, feared that contact with her baby—through "kangarooing," that is, skin-to-skin care—had caused the infection leading to its death next day. Her feeling of guilt persisted even though the HCPs and her husband tried to assuage it:

> Mother: You've got the feeling that everything went well until you're holding him for the first time. And then . . .
> Father: Yes, but there can be so many reasons for it. It's logical that you search [for reasons]. Especially as a mother, of course, you search for hundreds of things that you could have done better. And guilt . . .

The limited possibilities of direct contact meant that parents tried to engage with the child in other ways. For instance, they studied the baby's appearance or movements, that is, its breathing or skin color. Parents also explored other, substitutive ways of "doing something" for their baby and themselves. During the first NICU days, which were determined by physical distance to their baby, parents tried to find nonphysical ways of supporting and caring for their child, for example, talking, singing, blessing, or praying. Another couple sought to let their baby feel their positive attitude and love of life and tried to ease the seriousness of the situation by laughing and joking. Some HCPs did not recognize this as a coping strategy and were concerned whether the parents were aware of the severity of the situation. But most HCPs could frame the couple's attitude as a personal approach to dealing with the situation.

Providing Breast Milk

None of the mothers could breastfeed their baby. Pumping down breast milk and bringing it to the baby was perceived as one of the few possibilities of caring for the baby indirectly but physically. However, several mothers perceived the onset of lactation and the continuous pumping immediately after birth and during the baby's NICU stay as an additional source of stress and as something they felt urged to do by the HCPs for the benefit of the baby's well-being:

> [After birth-related general anaesthesia] I woke up, still clouded . . . and the midwife said that I have to pump now to stimulate my breast. I asked if this was really necessary. I couldn't even hold it [pump], my mother then held it. (Mother)

Mothers suffered severely from their breasts continuing to produce milk when their baby was dying, or after its death. It took some time for the anti-lactation medication to work:

> When we were waiting in this little room for them [HCPs] to tell us what's going on, the most horrible thing was . . . I noticed that I had to pump down milk. And while I was pumping down, they came in and told us that they couldn't do anything anymore. I thought: "Great! And what shall I do now with the milk?" I was completely . . . I then threw away the milk. (Mother)

Parents are not the Primary Caregivers

Instead of acting as primary caregivers, parents were busy with many administrative and organizational tasks amid the unexpected situation (e.g., caring for the baby's siblings, speaking to employers, etc.). They also faced many social encounters at the hospital and the mother's postpartum well-being. So besides profound concerns about their baby, parents experienced a co-occurrence of additional stressful situations. In general, preterm birth was a completely different situation to parents' previous associations with "parenthood." Apart from the genuine need for physical closeness to their baby, parents felt helpless toward its situation. They had to accept that their baby was receiving nonparental care. They depended on professional instructions about when, how, and for how long they could approach their baby. They relied on being offered the opportunity to be involved in care. They also depended on professional expertise for the appraisal of their baby's well-being. HCPs were used to the look of preterm babies, unlike parents, who had to

believe the professional caregivers that “everything is stable, it’s ok” (Father) even if the child’s appearance suggested otherwise. Although parents could call the NICU for updates at any time and experienced communication with the primary nurse or physician caring for their baby as good, this made them vulnerable to news from the hospital. Accordingly, parents experienced a lack of support in the absence of good communication with the primary caretaker or when they wished to have more information about the baby’s health from the baby’s primary HCP.

> You stand next to the incubator, thinking that it would be good if someone came by and told you how it [the baby’s condition] is. Perhaps I’m asking for too much, they don’t have the resources, the possibilities . . . They did tell us a bit that she’s doing ok. But [we would have needed] some more information. (Mother)

Parents with a medical or paramedical background oscillated between professional and parental perceptions of the situation. HCPs being unfamiliar with their dual background caused irritation among parents. One mother criticized the NICU nurses for realizing only at the very end that she was a nurse and for not involving her earlier in daily care. One father, a physician, blamed himself for not insisting more adamantly on a specific medical intervention for which he had expert knowledge when his son contracted a lethal infection. He felt that he failed to assume responsibility as a father with a medical background.

End-of-Life

In this second phase, it has become certain that the baby’s situation will no longer improve and that death is approaching. Despite the despair linked to this phase, this is where child–parent bonding can occur.

Caring for the Dying Baby

Parents were asked about how they wished to proceed when their baby was dying. These questions included, for example, the persons involved in caring for the dying baby and the setting in which the child would die. As mentioned, parents suffered from the limited possibility of touching their baby in the NICU. Most of them felt a profound need to fulfil this parental role and to give some warmth and support to their dying baby:

> The worst thing [about the incubator] was that you … can only stick in a finger after having it sanitized five times … Actually you have a huge

> desire to give some warmth to the baby and to feel him. And you can only do this when it's too late. Medically speaking it all makes sense … But this was something really beautiful as well as really sad. (Father)

Also, parents wanted to talk to their baby, express their feelings of gratitude or guilt, play some music, or sing a song. Some created such moments themselves; others were encouraged by the HCPs:

> They laid him into my arms … and just pulled the curtain. They instructed me like: "Tell him everything you may want him to know." And I sat there thinking, well … (laughs). This sounds so … I remember that I just sang a lot because I had always sung during pregnancy. And then I really sang one more time. (Mother)

However, parents also mentioned their struggle to accompany their dying baby and to hold him or her for the first and last time, even though all of them wanted their dying baby to be held. Some mothers needed support to overcome their initial hesitation to hold their dying baby, whereas others did not have the strength and asked their husbands to do so instead. For some mothers, it was important to give their husbands the opportunity to hold the child because they did not experience physical closeness during pregnancy. Even though some mothers were told by the HCPs that it is most beautiful for the baby to be held by the mother, they wanted their husbands to share this last experience with the living baby.

Creating Privacy for Dying

All parents were offered as much privacy as possible during the baby's dying process in the NICU. Some parents were separated from the open NICU space by a curtain drawn around the incubator. This separation provided parents with visual privacy but did not protect them from ambient noises and actions:

> It was weird because … we wanted to take leave [of our baby], but there were so many people fighting for another baby's life. I found this a bit awkward. When the curtain was closed, it felt ok. It was super-narrow, I think it [curtain] reached just around us … But it was closed. We still heard things, but then everything was gone. (Father)

Other parents were brought to a private room where they could take leave of their baby calmly and undisturbed. Parents could lie down on a bed with the baby. One couple experienced the transition to this room as very difficult because they could not hold the baby but first had to push it in a trolley across the corridor:

> Father: I remember that the walk down the hall was very burdensome.
> Mother: It was stupid because it [hall] is public space ... where every visitor can ... We had to put her into a little bed and push it [down the hall]. Of course we would have preferred to carry her.

Other parents criticized the clinical and functional design of the private room. HCPs, however, tried to create a pleasant atmosphere with the available options (e.g., by dimming the light or by adding some decorativeness or creating a homely atmosphere with a colourful cloth or a candle). Some parents reported a sense of ambivalence: They needed privacy on one hand, yet support on the other. One mother, for instance, felt an awkward tension between herself and the HCP in the room when she was crying in sorrow. One couple described how the physician stayed in the room without explanation.

Caring for the Deceased Baby

Retrospectively, all parents felt the need to spend some time with their deceased baby even though this varied from a few hours to a week. Some parents felt that they needed an extended period with the deceased body due to the incomplete pregnancy and the baby's short life span. They could continue to hold their baby for a while right after its death and could see their baby at any time, be it in the mother's room or in the room of quiet.

Parents positively recall those moments in which they could do what parents "normally" do:

> It felt very strange to [dress] such a cold child, it was . . . sad. But I think it helped us to take leave. (Father)

> It's almost schizophrenic, we were excited to dress her. Yes, we could at least do this, we couldn't do anything else. (Father)

Looking back, parents felt glad that the HCPs encouraged or even pushed them to spend time with their deceased baby in one way or another, even though it was something some parents initially felt hesitant about or wanted to push aside:

> I'm really glad that they consistently put me under pressure to take him. Because one memory about him is his weight: I took him out of the basket on the little cloth which was wrapped around him, and that's something which I felt long after: his weight. And this was so beautiful. (Mother)

> We wanted to leave but then a night nurse asked if we didn't want to dress her, which I'm really glad about. I then said no because I was afraid to hurt something like her little arm. She [nurse] then said that parents are usually doing this. And I was glad about that. Now, in retrospect: If she hadn't said this, we would simply have left. (Mother)

Some parents would have wished to be invited and encouraged more actively to hold, wash, or dress the deceased baby and to overcome their initial feeling of insecurity, hesitation, or paralysis. They regretted that the HCPs did not involve them more actively but instead accepted their initial reluctance. However, encouraging contact or touch to mourn the passing of the baby could also have an ambivalent effect if parents really did not want to spend time with their deceased baby and experienced the HCPs' insistence as directive and stressful. These parents wanted to preserve a certain memory of their baby and feared that it would negatively overlap with the image of the transformed dead body. However, other parents needed to witness this physical transformation from "lifeless" to even "more lifeless" (Father) to realize their baby's death.

The room of quiet was a place where parents could come to grieve for hours, days, or even years after losing their child. There, they felt connected not only with their child but also with other bereaved parents. Some mothers decided to keep their baby in their hospital room for several days to feel its presence, to become aware of its death, and to go through the process of farewell, even though some experienced this presence as ambivalent. None of the parents took their deceased baby home. One couple would have liked to, but they were not aware of this possibility.

Parents needed the certainty that someone would be caring for their child until the cremation or funeral. For instance, a mother who could not leave her baby alone in the room of quiet waited until an HCP found time to bring him to the pathology unit:

> I didn't want to let the baby lay there in such an exposed manner, [not knowing] who would enter the room, and for how long he would lay

> there. . . That was really heavy, I really cried a lot . . . I simply wanted to take leave but I sat there for 2.5 hours with my dead baby on my knees. (Mother)

It was also very important to the same mother that her child's body would suffer only minimal damage during the postmortem. She could not cope with the idea that her baby's body would be harmed and thus requested a limited biopsy, asking the pathologist to suture the child's body carefully and with dignity. Parents expressed their need to be considered a family even though their baby had died. One mother, for instance, appreciated that her primary nurse asked her how she and her dead baby, whom she kept in her room for a while, wished to be approached and talked to. Mothers were irritated if staff members did not know how to approach them and their dead child and just left without saying anything. In some rare cases, parents did not manage to separate from their baby and were told by the HCPs that the physical decay of the baby's body required them to bid farewell. All parents experienced final separation as very difficult as death became an irreversible reality and meant that they would return to their homes, which they had prepared for their baby's arrival, but without him or her.

Generating Family Memories

Midwives, nurses, and parents created memories of the living, dying, or deceased baby. They did so by making footprints, taking pictures, writing a diary (primary nurse on behalf of the parents), creating a stone (a memorabilia) for the baby, and by writing and drawing into a memory book available to all parents of deceased babies in the room of quiet. Generating memories was strongly encouraged by HCPs and retrospectively appreciated by all parents. Memories invoked by pictures helped parents to remember and visualize their baby's short life and their time together as a family. Even if some parents never looked at the pictures again, they still knew that these pictures existed. In most cases, parents did not take the pictures themselves due to their concentration on their dying baby and a lack of energy. Especially during the process of dying, parents initially found it awkward to be photographed:

> I found it weird that they photographed us during the "death scene." . . . Such a horrible situation, and she's taking pictures! I didn't understand it . . . In retrospect I'm glad that they did because it felt like a film. Like this you can really look at it again and also realize it. (Mother)

In retrospect, almost all parents appreciated the nurses' initiative. Only one couple did not want any pictures to be taken while their baby was alive, as they wished to spend the time remaining undisturbed. Even if parents used the time available differently, they all felt it was most precious:

> The five days, which were five regular days for others, were the five days of my daughter's life. (Mother)

Discussion

Our results have described how parents experienced the short trajectory of their extremely preterm babies from birth to death in a Swiss NICU. We have shown that this burdensome trajectory is linked to the transformation from assigned and distant parenthood to embodied parenthood and a sense of family bonding. For parents, this trajectory was affected by uncertainty, distress, inexperience, hopes, fears, and feelings of responsibility and guilt, all resulting in a liminal state (Wraight, McCoy et al. 2015). A lack of emotional preparedness, immediate separation, and the foreign NICU environment contributed to parents' perception of the situation as unreal. Liminality was experienced medically (uncertainty of diagnosis, prognosis, chances of survival), spatially (alienating NICU environment), socially (HCPs as primary experts, limited possibility of parenting activities), and emotionally/cognitively (detached state of mind, simultaneity of joy and sorrow). Parents experienced a compression of time and space between birth and death, during which occurred the process from assigned and distant parenthood to embodied parenthood (Figure 1). During the phase of uncertainty, when the baby received intensive care, parents experienced a series of challenges or obstacles to embodying parenthood. However, our study shows the importance of promoting bonding during the end-of-life phase so that parents overcome the "parental incompleteness" (Hutchinson, Spillett et al. 2012, p. 8) experienced in the phase of uncertainty.

Phase of Uncertainty

Due to their frailty, around half of the babies in our study were separated from their mother immediately after delivery and provided with neonatal intensive care if potential viability was assumed. This spatial separation (maternity ward and NICU) also symbolized two distinct focuses of care, which paralleled each other but were described as barely intertwined, as also reported by Flacking et al. (2006). In our study, the lacking interconnection was expressed, for instance, by

mothers feeling that their well-being mattered less to NICU HCPs, by the mothers' early discharge from hospital, and by a lack of rooming-in facilities for parents at the NICU. No explicit mention was ever made of the fathers' well-being. However, our results show a strong involvement of fathers in the NICU: for instance, as transmitters between the maternity ward and the NICU (information, but also transfer of expressed milk; Sweet 2008), as co-organizers of administrative tasks, as supporters of their wives, who suffered from birth- and NICU-related "shocks" and "films," and as active agents in terminal care if their wives lacked the necessary strength (for studies including the perspective of NICU fathers, see, for example, Mahan, Perez et al. 1981, Feeley, Sherrard et al. 2013, Garten and Hude 2014, Hasanpour, Sadeghi et al. 2016, Hynan 2016, Ionio, Colombo et al. 2016).

Despite separation after birth, all parents were supported to have immediate access to the NICU. As Hutchinson et al. (2012) pointed out in their qualitative study, "the initial visit to the NICU served as a confirmation that the parents actually did have a baby" (p. 15). Even though great effort was invested in facilitating parent–child encounter after birth, when facing their baby in the incubator, many parents expressed disbelief that this was "their baby" lying there. This incredulity was enhanced by the child's appearance, as other studies have also shown (Flacking, Ewald et al. 2006, Shin and White-Traut 2007, Hutchinson, Spillett et al. 2012). Despite immediate access, the interviewed parents had very limited possibilities of physical contact with their baby and thus had to find alternative ways of "doing something" for him or her. Accordingly, the limited possibilities of physical contact with the child (i.e., feeling, touching, and holding) were described as very strong parent–child bonding. These moments helped parents to grasp their baby's existence in the highly alienating NICU setting and gave them the opportunity to care for their baby physically. However, holding and touching were related to feelings of extreme insecurity, helplessness, frailty, sensitivity, responsibility, and guilt, as exemplified in the case of a baby who died 1 day after kangarooing for the first time. Physical contact could occur neither intuitively nor "naturally." Rather, HCPs decided on parents' access to and contact with the baby and supported and instructed parents in caring for him or her (Aagaard and Hall 2008, Fenwick, Barclay et al. 2008). HCPs also restricted access to the child during specific medical interventions. As our study and others show, parents who were not allowed to visit their baby due to unscheduled or extended clinical interventions were in need of explanations to prevent them from worrying (Guillaume et al., 2013).

Evidence shows that NICU parents wish to do things that parents of newborns "normally" do, "such as holding, comforting, and feeding" (Currie, Christian, et al., 2016, p. 4). Also, "parents need and want to be given opportunities in which they can act as normal parents" (Fenwick, Barclay et al. 2008, p. 49, see also Kearvell and Grant 2010). The provision of breast milk—in the cases analysed in our study through milk expression rather than direct breastfeeding—was one of the few ways in which mothers could physically "do" something for their baby and experience maternal "agency" (Sweet 2008, Ikonen, Paavilainen et al. 2015). Due to its known benefits for infants in general, and extremely preterms in particular, breast milk is the nutritional gold standard in NICUs (Cricco-Lizza 2014, Spatz and Edwards 2016). Cricco-Lizza (2011) showed that nurses are highly motivated in "[m]aximizing babies' potentials in the midst of uncertainty" (p. 401) and devoted to infant well-being. Their aim is to enhance the chances of a good outcome. Ensuring the provision of breast milk is one way of demonstrating this commitment. Some studies on lactation and breastfeeding in the NICU focus on the reciprocal benefit for baby and mother (Bonet, Forcella et al. 2015, Kavanaugh, Roscigno et al. 2015, Shattnawi 2015) and consider it an "important rit[e] of passage into motherhood" (Shattnawi, 2015, p. 81).

But matters might be different if breastfeeding is not yet possible. Various studies, including our own, indicate that some mothers experienced a strong ambivalence when providing breast milk by means of expression. Among the reported reasons were lack of professional support, lack of knowledge on the benefits of breast milk, the perceived coldness of pumping versus the intimacy of breastfeeding, the stressful situation in which mothers have to produce enough milk, the short-term burden versus the long-term benefits, and physical pain (e.g., sore nipples) (Cricco-Lizza 2014, Lucas, Paquette et al. 2014, Ikonen, Paavilainen et al. 2015). Mothers who perceived the provision of milk as difficult held on to it because they were told about the nutritional and immunological benefits for the baby. They felt a duty to enhance their infant's outcome. Referring to Ikonen et al. (2015) emphasized "that expressing required perseverance, resilience, and motivation" and "interfered with everyday life" (p. 402). However, as Flacking et al. (2006) showed in their literature review, "successful" breastfeeding also involves other factors, such as "maternal enjoyment, attainment of the desired maternal role and life-style compatibility" (p. 71). Studies illustrated how the provision of breast milk in the NICU is strongly linked to the idea of being a "good mother" (Ikonen et al., 2015, p.

401; Shattnawi, 2015, p. 75). Vice versa, if this is not possible, or if the milk is rejected, this entails a feeling of failure and guilt (Sweet, 2008).

Our study shows that this ambivalence is mirrored in the normative pressure and stress perceived by some mothers with regard to the provision of milk. In their review, Ikonen et al. (2015) described that in some of the cases they examined, "expression was approached with perseverance, thereby causing exhaustion in the mothers" (p. 398). Evidence suggests that the constant concern with pumping down milk for the baby may enhance mothers' distress and even "their experience of liminality" (Shattnawi, 2015, p. 81). The stressful onset immediately after birth and the continuation of lactation beyond the baby's death are powerful images of the emotional and physical struggle undergone by mothers. Thus, for HCPs, this involves walking a tightrope between encouraging mothers and considering their well-being on one hand, and encouraging them to provide breast milk as soon as possible after delivery on the other (Bonet, Forcella et al. 2015).

However, we have to keep in mind the retrospective design of our study. All mothers who experienced pumping as burdensome lost their infants. To them, pumping was "useless," as one mother put it. However, they probably would have interpreted pumping—and refraining from pumping—differently if their infant had survived. Sweet (2008) and Lucas et al. (2014) found that mothers motivate themselves for burdensome pumping because it is a precondition for later breastfeeding. Our mothers, however, interpret pumping as negative because they never reached this future.

End-of-Life

After the decision to withdraw further intensive care was made, parents were offered a private space where they could be with their child during its dying. For the parents under study, this was the first time that they could be alone with their child. Parents described two ways of how privacy was offered: first, by pulling a curtain around them and their child; second, by transferring the family to a private room. Even though this transfer is very short, parents described it as very difficult. Hall and Brinchmann (2009) explained how sensitive NICU parents react to spaces and to transfers across spaces. Once the room in which the child would die was defined, parents in our study expressed a strong desire to experience intimacy with their child. But they also needed to feel certain that quick professional support was available if needed. Parents expressed irritation when professionals either disturbed these intimate moments or when professionals were not available.

The parents in our study all lost their babies after a short NICU stay. For them, holding did not represent stabilized or increased well-being but the child's deteriorated health condition and the onset of dying. They experienced physical contact with their baby only during and after dying and considered it both a gift and a parental duty. Even though most mothers in our study wanted to hold their dying baby, others either did not feel strong enough to hold the baby or wanted their husbands to experience this very last moment of closeness. Thus, although all mothers wanted their dying baby to be held, not all of them could or wanted to do it themselves (McHaffie, Laing et al. 2001, Moro, Kavanaugh et al. 2011). During observation, we heard about a few cases where parents did not stay with their dying child, a decision that caused profound irritation among HCPs. Holding the dying baby, or "skin-to-skin care" between dying babies and their parents, is underinvestigated yet considered a strongly recommended caring practice, as a Scandinavian study shows (Kymre and Bondas 2013). Skin-to-skin care is based on the assumption that it provides "mutual proximity and comfort for dying preterm newborns and their parents" (Kymre & Bondas, 2013, p. 671). Thus, "closeness, touch and comfort" (Kymre & Bondas, 2013, p. 672) is encouraged not only for the dying baby's well-being but also for parents' current and future well-being. Baby-parent proximity is thus considered a criterion of a "good" NICU death.

Our study shows that parental agency does not cease with the baby's death. Rather, most parents wished to spend some time with their deceased child during which they sensed his or her presence or prepared him or her for definite physical separation. Relevant studies emphasize the importance for parents to continue parenting also after the baby's death (Currie, Christian et al. 2016) and to have the "opportunity to spend time with their baby as a member of the family, without all the tubes and wires" (Kymre and Bondas 2013, p. 670). Our data showed that parents co-created a sense of family by spending time together and by photographically documenting and thus visibly representing themselves as a family unit (Cortezzo, Sanders et al. 2015). Parents either created memories themselves, or were assisted by HCPs with collecting memories, or HCPs created memories of their own accord and gave the photographs to the parents at hospital discharge. Even though many parents initially felt awkward about being photographed during their baby's dying, retrospectively they highly appreciated these tangible memories, as confirmed by other studies (Lundqvist et al., 2002). Henley and Schott (2008) found that these "[p]hysical items connected with their [parents'] baby may help to confirm the reality of his or her short existence and provide comfort as well as a focus for their grief in the longer term" (p. 327).

On balance, the parents in our study experienced intensive embodied parenthood mainly during the process of dying and after death. Despite professional support and suggestions, they could then enter into direct, mostly non-moderated interaction with their child. Thus, in this final stage, they could do “things parents normally do” (e.g., have the baby close, give him or her some warmth during dying, wash, dress, and hold him or her during and after death). Along with these parenting activities, parents experienced closeness or guilt and felt deeply responsible for protecting their deceased baby from exposure or damage and for safeguarding his or her dignity (Sadeghi, Hasanpour et al. 2016). Thus, for the parents, the baby’s vulnerability and the duty of “caring” for and protecting him or her continued postmortem. This enabled them to regain parental agency after being observers and onlookers and to experience privacy after being observed and watched.

Professional Support

Our results show that parents find themselves in extraordinary distress during the dying trajectory of their extremely preterm infant and thus depend on professional support for the transition from assigned and distant to embodied parenthood. However, support in caring for the dying and deceased babies was described as a tightrope walk between professional encouragement and normative pressure. Most parents appreciated the strong support of HCPs in retrospect or regretted not being encouraged more strongly. Only a few parents perceived a negative normative pressure by the HCPs. Even though HCPs’ reference to “what other parents do” was appreciated by most parents, this may refer to ideas of “good” NICU parents. Barlow and Chapin (2010, p. 326) described that in the NICU cultural constructions of a good mother as the one who “promotes the well-being and development of her children and is almost always patient, protective, nurturing, and generous” are not applicable. However, our results indicate that expectations existed about what NICU mothers should do and that tensions arose when mothers did not fulfil or struggled to fulfil those expectations. Among these were the perceived prioritizing of the baby’s well-being over the mother’s, the provision of breast milk, the presence in the NICU, and holding the dying or deceased baby despite physical and mental distress.

Furthermore, parents reacted sensitively to how HCPs approached and communicated with them and with their dying or deceased child. Our results show that the continuation of embodied parenthood and family construction after the baby’s death calls for highly aware and sensitive verbal and nonverbal communication. For example, Henley and Schott (2008) emphasized the

importance of choosing a sensitive language when approaching bereaved parents, avoiding clinical terms, and instead talking about the baby in a person-like manner respecting his or her gender and name. The construction of embodied parenthood in the NICU thus involves viewing and treating the deceased baby as a person, and parents and the baby as a family. These are among the central concepts of neonatal palliative care as described by Stafford (2015) and Ahern (2013). They are based on focusing care on the child–parent dyad (Fenwick et al., 2008), respectively on the "family-professional partnership" (Kenner, Press et al. 2015, p. S20). Thus, in addition to comfort care for the dying child, this involves communicating palliative care options to parents, creating moments and spaces for parents and children to experience themselves as families, appreciating the child's life, creating memories, acknowledging different manifestations of grief, and supporting parents to let their child go (Ahern 2013, Stafford 2015, Abraham, Battaglia et al. 2016). Our results illustrate the necessity of further institutionalizing multiprofessional palliative and bereavement care concepts in neonatology units to enable "quality care" for the babies and "grief support" for the parents (Kenner et al., 2015, p. S19) and HCPs (Garten and Hude 2014). Ideally, such care is not limited to the hospital stay but is initiated—in the case of anticipated extreme prematurity—before hospital admission and continued after hospital discharge.

Conclusion

In conclusion, our study illustrates that parents of extremely preterm babies suffered from unpreparedness of becoming parents: They were considered parents (assigned parenthood) with the birth of their baby, but to actually feel like parents they needed to go through a process of biological and psychosocial bonding. In our sample, child–parent embodiment through holding, touching, smelling, caring, and protecting barely occurred before dying (distant parenthood). Thus, caring for their dying and deceased child enabled parents to become parents in an embodied sense. Even though this trajectory can be very short in time, it is of crucial importance that parents receive the professional support needed to experience embodied parenthood which is grounded on palliative and bereavement care concepts elaborated for neonatology.

Conflicts of Interest

The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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End-Of-Life Decision Making for Parents of Babies Born Extremely Premature

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Author Contributions

All authors contributed to the different parts of the study. All authors designed the study and drafted the interview guide. Mrs. Hendriks and Dr. Abraham carried out the data collection and analysis. Both authors interpreted the data and contributed to the initial manuscript. All authors revised and approved the final manuscript. All authors have participated sufficiently in the work to take public responsibility for appropriate portions of the content.

Additional members of the NRP67 Neonatal End-of-Life Study Group include the following: Jean-Claude Fauchère, Hans-Ulrich Bucher, Ruth Baumann-Hölzle, and Thomas Berger.

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Abstract

Objective: To explore parental attitudes and values in the end-of-life decision-making process of extremely low gestational age newborns (gestational age <28 weeks).

Design: Hermeneutically oriented qualitative research design with in-depth interviews.

Setting: Level III NICU in Switzerland.

Participants: Purposive sample of seven couples, five mothers, and one father (20 parents).

Methods: Qualitative content analysis was used to categorize and interpret themes from parents' narratives.

Results: Parents described factors that affected the decision-making process in satisfactory or unsatisfactory ways. Transparent information, empathy, and honesty enhanced communication between parents and the health care team. Lack of transparent information and continuous support decreased satisfaction. The level of involvement in decisions differed by setting. Most parents made decisions regarding lung maturation and/or initiation of care in the delivery room. Parent participation in the NICU was experienced differently. Contrary to the hospital's ethical model, few parents recalled being involved in the decision-making process. Some parents experienced a dissociative state of mind that hindered their involvement, whereas others felt actively involved.

Conclusions: Our results suggest the need for careful and continuous professional evaluation of parents' wishes about involvement in the decision-making process along with describing medical facts and treatment options. A lack of attentive listening and dialogue may cause paternalistic decision trajectories.

Keywords: extremely preterm infants; parental involvement; end-of-life; shared decision-making; qualitative research

Introduction

The birth of an extremely preterm infant (gestational age < 28 weeks) often comes unexpectedly. Decisions, then, must often be made under circumstances that can be emotionally stressful or morally burdensome for parents and/or health care professionals (HCPs) (Provenzi, Barello et al. 2016). Because infants born at the borderline of viability have a wide range of outcomes, it is hard to predict, before of after birth, whether an infant will die or survive with or without impairment. Prognostic uncertainty make decisions about life saving treatment, particularly, complex (Leuthner 2014).

Over the years, different decision-making approaches have been developed to guide parents through the uncertain context of prematurity. In the past, HCPs considered it their professional prerogative to make decisions about initiating or withholding intensive care treatment. This concept is known as paternalism. In recent decades, however, the focus of decision making has shifted to include concepts such as informed decision making, informed shared decision making, partnership, patient involvement, patient-centred care, and evidence-based patient choice (Moumjid, Gafni et al. 2007). Currently, most guidelines and policies advocate shared decision-making and promote parental involvement before and after birth. They are based on the premise that decision-making should be a collaborative venture between neonatal HCPs and parents in making decisions about their infant's care (ACOG 2015). In fact, it has been shown that parents want to participate in making life-support decisions about their infants' care (Moro, Kavanaugh et al. 2011, Provenzi, Barello et al. 2016, Weiss, Barg et al. 2016). Moreover, parental involvement in combination with compassionate communication, consistent information, and support from HCPs has contributed to parental satisfaction with decisions (Brosig, Pierucci et al. 2007, Obeidat, Bond et al. 2009). Hence, the relationships and communications between HCPs and parents are key in establishing parental involvement (McHaffie, Laing et al. 2001, Moro, Kavanaugh et al. 2011), whereas nurses play important roles in supporting parents in making decisions (Kavanaugh, Moro et al. 2010).

However, despite the prevalence of this perspective, implementation of shared decision-making approach in practice has remained difficult (Stiggelbout, Van der Weijden et al. 2012, de Vos, Bos et al. 2015). Consequently, empirical data are important to provide further insight into how parents of extremely preterm infants experience communication with the health care team and how this affects involvement in end-of-life (EoL) decision making. Therefore, we examined how parents reach medical decisions for their extremely preterm infants in our perinatal centre in

Switzerland. We sought to identify communication patterns between parents and neonatal HCPs that lead to the perception of a satisfactory or unsatisfactory decision process. We further assessed the degree of parental involvement within the ethical decision-making model applied in our perinatal centre between 1994 and 2015. This study was part of a larger research project on EoL decisions for extremely preterm infants in Switzerland. In this qualitative study we aimed to shed light on the perspectives of parents, whereas the quantitative part of our project focused on the perspectives of society and of HCPs who work in Level III NICUs (Hendriks, Bucher et al. 2017, Hendriks, Klein et al. 2017).

Methods

Sample and Setting

Out of 42 potential cases, 20 parents of 13 extremely preterm infants (7 couples, 1 father, and 5 mothers) were recruited by purposive sampling. Potential participants who met the following inclusion criteria were recruited: parents of newborns who were born alive at less than 28 weeks gestation and died in the delivery room or in the NICU from 2013 through 2015. Parents with various linguistic backgrounds were invited to participate by means of a translator. A timeframe of 1 to 2 years between the infant's death and the interview was chosen in recognition of the mourning process and of parents' vulnerability (Rosenblatt 1995, Caeymaex, Speranza et al. 2011). To reach a homogenous sample, participants were excluded if they gave birth to two or more infants (i.e., multiple births) or if their infants were stillborn. Participants received a letter (written in German and English) from an attending neonatologist and the former director of the neonatology department to solicit participation in the study. Interested participants were asked to contact the interviewers directly by e-mail, phone, fax, or letter. The Ethics Committee of the Canton Zurich (Switzerland) approved the study protocol. Participants gave informed written consent for the research use of the collected data.

Data Collection

Data were collected through narrative interviews with semi-structured follow-up questions in order to clarify specific themes (see Table 1). The interviews were used to explore participants' experiences of prematurity, communication with the health care staff, and the EoL process. In developing the interview guide, the literature on EoL decision-making for parents of extremely

preterm infants was taken into account (Rosenthal and Nolan 2013, Leuthner 2014). Additionally, this was complemented by our field notes from observations in the NICU, informal interviews with various staff members (i.e., neonatologists, a physiotherapist, a psychologist, a pastor, and a music therapist), and chat forums used by affected parents. The interviews started with a narration of the participants' experiences, i.e. they were invited to share their stories starting with the pregnancy until after the death of the newborns. Some participants showed photographs, diaries, and other memories of their newborns. The interviews were conducted in a place selected by the participants. The interviews were audio recorded, lasted approximately 60 to 160 minutes (97 minutes on average), and were transcribed by a professional transcriptionist.

Table 1. Overview of Interview Guide

Main Areas	Example Questions
Narration of parents experiences	Please tell me about your baby.
	Parents are (non)verbally encouraged by the interviewers; 'Hmm', 'yes', 'I see', 'What happened before/after/then?'
Communication with HCPs	How did you experience (verbal and/or non-verbal) communication with HCPs?
	What were the main obstacles?
Parents' experiences with EoL decisions	What were the main events leading up to EoL?
	How did you experience the EoL decisions to be taken?
	What were expected and/or unexpected EoL decisions that had to be taken?
	Who were the main actors involved in decision-making?
Parents' wishes and preferences	What were your wishes and preferences in caring for your infant?
	What would you recommend other parents who are in a similar situation?

Data Analysis

Data analysis followed hermeneutically-oriented, qualitative content analysis, which takes into account previously defined research questions outlined by the literature and allows categories to

emerge out of the data. This deductive-inductive procedure was guided by Kuckartz's approach of content analysis, which integrates elements from grounded theory such as theoretical memos and iteration to generate not only descriptive results but also conceptual models of the topics under study (Kuckartz 2014). Hence, this approach is focused on the importance of context in determining meaning that is data-driven and iterative. Our analysis included the following steps: (a) inductive-deductive generation of thematic categories, (b) first rough coding, (c) further development and differentiation of categories on the material, (d) coding of all transcripts and (e) category-based analysis and writing of the research report (Kuckartz 2014).

The coding process involved a search for commonalities among the transcripts through an iterative process. First, an initial coding scheme was developed based on our research question, interview guide, reviewed literature, and the field notes from our NICU observations. Second, both authors coded the first four transcripts to achieve congruent coding practice using the qualitative data analysis software Max Qualitative Data Analysis (MaxQDA version 12). Third, the coding scheme was refined with codes that emerged from the collected data. It comprised a chronologic axis (i.e., pregnancy, birth, liminal phase, EoL, dying, and death) and a thematic axis (i.e., experience, values and attitudes, support, and communication). Fourth, transcripts were randomly divided between the two authors and coded along the coding scheme. Similar codes were summarized and clustered, whereas interpretative notes were formulated as theoretical memos. Finally, the clustered codes were grouped (i.e., systematized) for in-depth analysis of specific themes in the data (Silverman 2015). On one hand, this resulted in a comprehensive, descriptive running text. On the other hand, we gradually moved from description to the generation of a conceptual model of the main topic under study (i.e., decision-making).

Several strategies were used to ensure accuracy in the data analysis. First, credibility was complemented by peer debriefing among the authors. All codings, summaries, and memos were cross-checked and complemented by the other researcher. Discrepancies were resolved through repeated discussion. Second, validation and feedback were sought by means of interviewing five HCPs who were not involved in the participants' cases. This was done to obtain further insight into the experiences of participants.

Results

Sample Characteristics

Participants included seven couples, five mothers, and one father. Their ages ranged from 33 to 46 years at the time of participation (see Table 2). The extreme preterm infants were born between 22 and 27 weeks gestation, and birth weight ranged from 340 to 1100 grams (see Table 3). There were five live-born infants born at the limit of viability for whom intensive care was withheld because of medical indication or parental decision; these newborns received comfort care. Resuscitation was attempted for eight newborns, and seven newborns were admitted to the NICU.

Table 2. Parental Characteristics

Characteristics		**N**
Sex	Male	8
	Female	12
Age	25-30	1
	30-35	5
	35-40	8
	40-45	4
	45-50	2
Cultural Background	Swiss-German	13
	German	3
	Italian	2
	Turkish	1
	Serbo-Croatian	1
Religion	Christian	18
	Jewish	1
	Muslim	1

Table 3. Infant Characteristics

Characteristics		**N**
Sex	Male	7
	Female	6
Age	22 0/7-6/7	1
	23 0/7-6/7	1
	24 0/7-6/7	5
	25 0/7-6/7	4
	26 0/7-6/7	-
	27 0/7-6/7	2
Weight in Grams	250-500	3
	500-750	6
	750-1000	2
	>1000	1
Number of Survived Hours/ Days	Hours	6
	<5 days	2
	5-10 days	3
	10-15 days	2
Cause of Death	Extreme Prematurity	5
	Intra-uterine growth restriction	1
	Respiratory Disease	3
	Necrotizing Enterocolitis	1
	Major IVH	3
Place of Death	Delivery Room	6
	NICU	7

Ethical Decision-Making Model

The HCPs we interviewed indicated that from 1994 until 2015, the University Hospital applied an ethical decision-making model in which the neonatal HCP involved parents by interpreting their values and informing them of the best possible treatment determined by the neonatal team. Ethical discussions were initiated for all extremely preterm infants born before 26 weeks gestation, newborns with severe malformations, and newborns with poor neurologic prognosis. After a structured discussion, the neonatal staff discussed at least three different treatment options and deliberated until consensus on the best treatment strategy was reached. The team took into account the newborn's best interests and the parents' values. After that, the physician and nurse dedicated for the support of the parents would inform parents about the infant's condition, the prognosis, and the health care team's treatment recommendations. Parents were asked for their informed consent for the proposed treatment and were also informed of their veto right. If parents disagreed with the recommended treatment, time was given to allow for further discussion with the neonatal team. With the exception of emergency resuscitation, the norm was that all ethical discussions included parents in the final decision and its implementation.

Communication: To Be Treated With Honesty, Sympathy, and Transparency

We found that some elements enhanced and some challenged communication between the participants and the health care team and resulted in the perception of a satisfactory or unsatisfactory decision process. One crucial factor was a thorough medical explanation of extreme prematurity, including the chance of survival, possible complications or disabilities, and projected outcomes in early childhood. This information enabled the participants to prepare for the process ahead. Participants perceived unsatisfactory decision-making because of communication challenges. At the prenatal stage, parents reported that they were not always informed about the hospital's gestational cutoff policy, i.e., the non-initiation of treatment for infant's born before 24 weeks gestation. Some parents received only implicit or minimal information on the hospital's guidelines, which prompted them to actively seek full and explicit information. Other parents did not receive any type of prenatal information, and no transparent reasons for why treatment was withheld at the prenatal stage were given:

> We learned about this [the gestational age cut-off] later. He was 23 weeks and some days. So that was the aggravating part of the story, so

> shortly before, why? … And that takes a while before you can accept it. – Mother

Other parents experienced dissatisfaction in the NICU because of lack of continuity of parental support and poor communication. One mother described how communication depended on which nurse was present: “One would inform us well and would start telling us everything on her own; with others you had to drag the information out of them.”

Parents greatly appreciated when HCPs explained matters; offered the chance to ask further questions; and showed empathy, experience, familiarity, continuity of care, and honesty. Parents wanted to be treated like “a normal human being.” One father reported that the physician had a professional expertise and a “human side; she simply listened to us and was a very sympathetic person.” In addition, non-verbal communication was valued. Thus, professional support did not merely involve informing parents about the risks of premature birth. It also meant listening to parents’ concerns and being there for them around the clock:

> One of the nurses even wrote me an e-mail in the middle of the night to tell me that my baby was sleeping well. That touched me so because […] that was my first night at home. I found that amazing. What a calling, such a job. – Mother

These tasks were not limited to HCPs. Other staff members such as the spiritual caregiver or psychologist were also available to support the family.

Parental Involvement in End-Of-Life Decision-Making

Although decision-making before birth is only possible when there is time, a majority of parents were able to consider the option of steroids (for the maturation of the fetal lungs) and/or make decisions regarding resuscitation or initiation of intensive care. Parents described their active involvement in decisions to withhold or initiate care before birth: “We had the option not to do anything and then she would die after a while. Or we had the option to try everything possible. […] We discussed this and we said we would try everything." Parents whose infants died in the delivery room also wanted everything possible to be done to enhance the potential for survival; however, for some infants, care could not be initiated because their prematurity. A few parents decided not to initiate care for their infants in the delivery room to prevent them from suffering.

Furthermore, parents experienced involvement in decision-making in the NICU in different ways. Most parents did not feel actively involved, and some experienced a dissociative state of

mind that hindered their involvement. Few parents felt actively involved, and an overwhelming majority felt as if everything was already decided. Parents explained that the health care team informed them that "nothing more could be done." Parents therefore did not feel that they had taken part in decision-making:

> What was really to decide? But actually […] we relied on the staff and their advice. They have more experience, when they say it does not make any sense, then that was the decision. There was not really anything to decide. – Father

From their perspectives, the decision was clearly determined based on medical facts and limits. In fact, parents did not feel that real decisions needed to be made. One father had "a huge trust in the entire [health care] team and knew they tried everything." Parents felt comfortable with the decision to withdraw care based on their trusting relationships with the attending physicians and were convinced that the health care team tried everything. However, some parents were dissatisfied with their level of involvement and felt that the options and decisions presented by the health care team might have been biased and directive:

> The doctors make strong suggestions so that you cannot really decide […] you do what the hospital offers you. A different hospital might act differently […] possibly there were other options, but when you do not know what other options there are, how should you decide? – Father

A second group of parents reported that they experienced neither non-involvement nor involvement. Their minds were clouded by consuming shock, which made it difficult, and for some parents impossible, to be actively involved. For example, some parents said that decision-making was not a priority. Their dissociative state of mind left them unaware or incapable of participating in a decision-making process:

> I so to speak just watched as if I was not involved […] the entire time, I was personally affected but I did not experience it that way. It was like watching a movie. I was not really aware, and I could not really perceive the situation. It was as if in a dream, it could not be true, everything was fine. It took a while before I completely understood that it was my child, my child which was dying. - Mother

Finally, some parents recalled that they expressed their personal preferences and wishes to the health care team and subsequently were involved in decision-making. For instance, they considered it most important in terms of their parental responsibility and well-being that their newborns would not experience pain. Some parents explicitly opposed prolonging suffering with experimental treatment. One mother felt empowered by her decision to initiate intensive care treatment after feeling helpless and passive in the antenatal ward. But when faced with the decision whether or not intensive care should cease, this active decision-making role was challenging:

> The doctors thought I should decide when to take out the [breathing] tube. That is not an easy decision, not at all. I simply believed he should not suffer so long because of me. You could really notice he was slowly closing his eyes. – Mother

Parents appreciated the ability to make decisions without the direction of health care experts, but this also was difficult for some parents because of the emotional nature of the situation. In retrospect, however, parents appreciated and valued their involvement and decision-making was perceived as an act of parental responsibility:

> I did not experience this moment as a freedom but rather as a responsibility of course because this baby cannot decide for herself. We are her parents and we should make this decision. And we should decide what is best for our baby. Now in retrospect, I regard that as a great act of love. But in those hours, I thought I would die. But you do not die and you go on and you have to decide. – Mother

Discussion

We describe the decision-making process of parents whose extremely premature infants died in the delivery room or NICU. Our study confirms that parents of extremely preterm infants need specific and continuous support, in which high quality communication with the health care team plays a crucial role (Wigert, Dellenmark Blom et al. 2014). Consequently, parents in our study depended on the willingness of HCPs to provide that information. Clear and transparent medical information along with emotional support from caregivers and other NICU staff helped parents to cope and supported their decision-making. Additionally, favourable communication facilitated parents' confidence in recommendations for treatment. Lack of continuity and transparency in communication challenged the trusting relationship with the entire health care team. Poor

communication was previously considered an indicator of non-involvement in decision-making (Boss, Donohue et al. 2016). However, it is not sufficient to empower parents and involve them in decisions without sensitive communication. This can result in pressure that makes some parents feel as if they must participate. Hence, parental participation and involvement are affected by and depended on high quality communication (see Figure 1).

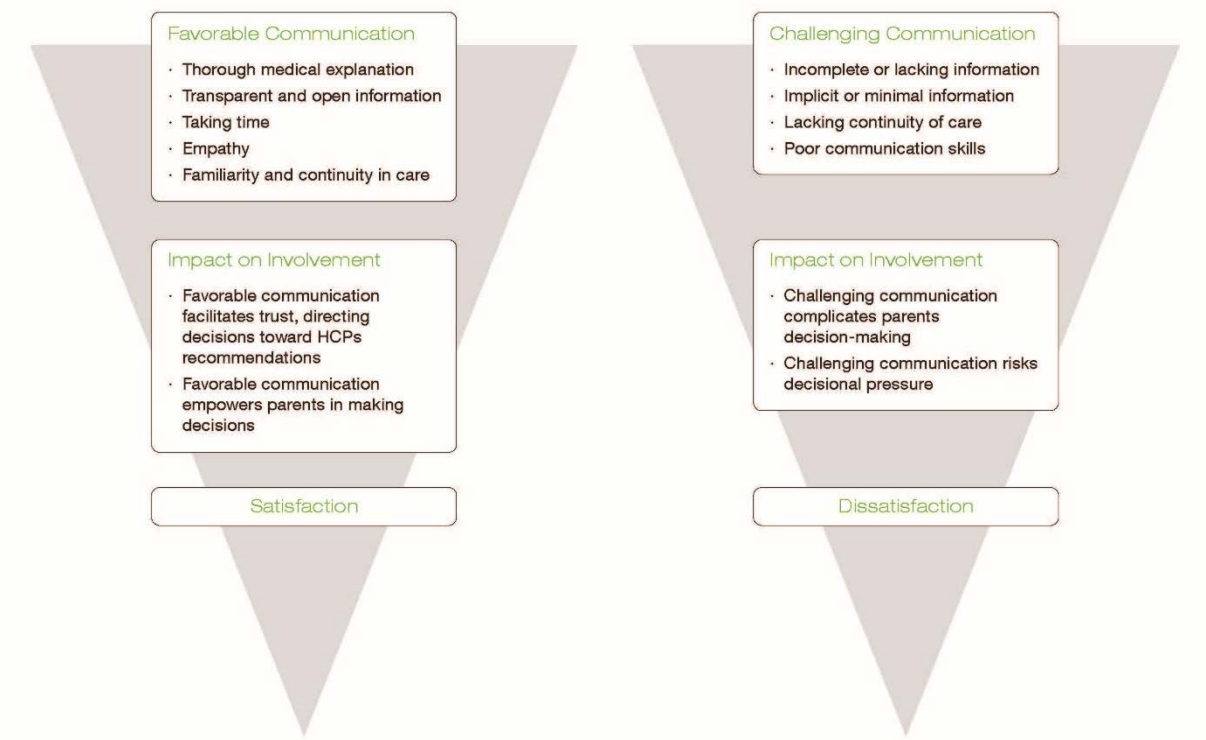

Figure 1. Elements that favor or challenge communication in the decision process

Unclear or inadequate communication about treatment options during the prenatal and antenatal periods was one of the most important factors to cause friction between parents and HCPs. Particularly, parents commented on the hospital's policy to limit the initiation of neonatal intensive care for extreme premature infants to a certain gestational age. At the time of our study, this limit was 24 0/7 weeks gestation in accordance with the prevailing national guidelines (Berger, Bernet et al. 2011). Thus, infants born before 24 0/7 weeks gestation were usually not considered eligible for life-sustaining treatment and resuscitation. A pressing question is whether parents should be informed about these considerations. The parents who participated in our study expressed the wish for transparent information and explanation of the ethical and medical arguments for withholding

life-sustaining treatment and resuscitation. Some parents retrospectively found out or considered that their newborns had nearly reached the limit of viability. This left them feeling confused and might have complicated the grieving process. Additionally, care for the most immature infants is not only evidence-based but also strongly influenced by the local NICU culture and national policies. In fact, infants born between 23 0/7 and 23 6/7 weeks gestation are provided treatment in some hospitals but not others (Guillen, Weiss et al. 2015). Whether, when, and how treatment divergences should be discussed with parents is a question yet to be resolved that warrants further extensive ethical and medical discussion.

The main purpose of our study was to assess parental involvement in the decision-making process. We found that the level of involvement in decisions seemed to differ between the delivery room and the NICU. Decisions regarding lung maturation, resuscitation, and/or initiation of care were thoroughly discussed and most parents wanted “everything done.” In the NICU few parents in our study recalled being actively involved in the decision-making process.

Similarly, in a study on parental EoL experiences in Switzerland, Zimmermann, Bergstraesser et al. (2016) found that decisions in the NICU were significantly less often shared between parents and the health care team than decisions in other paediatric EoL cases These findings stand in contrast to the hospital’s ethical decision-making model, the purpose of which is to provide parents with full information about the condition of the newborn, the prognosis, the different therapeutic possibilities, and the recommended treatment approach (Arlettaz, Mieth et al. 2005). If the parents do not agree with the recommendations of the team, a new ethical round including the parents takes place (Baumann-Holzle, Maffezzoni et al. 2005). In an evaluation study on EoL decision-making conducted by HCPs in the same hospital 10 years earlier, the authors found that 92% of parents were actively involved in decisions to withdraw care (Arlettaz, Mieth et al. 2005). This discrepancy between the ethical protocol and the perceptions of parents regarding EoL discussions can be explained in four different ways.

First, in retrospect parents might have not have experienced their own involvement as a deliberate act. Most parents described their trust in the health care team and valued physician recommendations and medical expertise. Parents might have preferred to delegate decision-making to the physician, and investigators found that some parents favoured a passive role (Weiss, Barg et al. 2016). Although most parents would like some involvement in decision-making, only a few sought full responsibility (Boss, Donohue et al. 2012). It is suggested that parental involvement should be tailored to individual preferences and needs (Madrigal, Carroll et al. 2012, de Vos, Bos

et al. 2015). A problem lies, however, in the fact that such an approach requires a deliberate, decision-making process (Moumjid, Gafni et al. 2007). Even though the hospital's ethical decision-making model takes into consideration parents' values and aims to involve parents in final decisions (Baumann-Holzle, Maffezzoni et al. 2005), this type of involvement has been criticized as not being a shared collaboration (Légaré and Thompson-Leduc 2014). A shared approach should involve the exchange of information between HCPs and parents, deliberation about all treatment possibilities, and joint decision about which treatment should be implemented (Légaré and Witteman 2013). This brings us to our second point.

Parents might not have had the impression of being involved in the decision-making process. Information was presented one-directionally, and the physician recommended one treatment option. This could potentially lead to directive counselling in which the physician's view swayed the parents. Researchers have confirmed that HCPs can steer decisions toward a specific treatment without outlining or explaining alternatives (Haward, Murphy et al. 2008, Moro, Kavanaugh et al. 2011, Stiggelbout, Van der Weijden et al. 2012).

Another explanation might be that parents retrospectively considered their non-involvement as unsatisfactory to better cope with the situation and the EoL process in the long term. This might be because paternalism has a long tradition in the Swiss health care context. In general, Swiss HCPs acknowledge the importance of shared decision-making, but the level of patient involvement still varies (Cornuz, Kuenzi et al. 2011). Additionally, Swiss guidelines on extreme preterm infants are based primarily on HCPs' assessments (Berger, Bernet et al. 2011). Consequently, these features might have resulted in a setting in which parents continuously relied more heavily on the physician's judgment.

Finally, shared decision-making in the ethical model might have been confused with the more broadly defined patient-centred care or informed consent. Neonatal HCPs may already partly engage parents, but this should also include the ethical and moral standards of a shared approach. To begin with, a shared approach should be provided to all parents with room for adaptation so that the level of involvement corresponds with their individual abilities and interests (Légaré and Witteman 2013). With a shared approach, parents are not abandoned to make decisions alone, rather they are empowered to optimize their expertise in a supportive environment (Légaré and Thompson-Leduc 2014).

Our findings indicate that parents preferred a shared approach with their neonatal physicians rather than a paternalistic or informed approach. When time allowed, most parents in our study actively and deliberatively expressed a clear preference regarding the course of treatment in the delivery room. Skilled communication in the NICU to involve and inform parents is an essential part of and the first step to ethical decision-making. Moral deliberation includes reflection on aspects such as the child's best interests, the values of the parents, and the harms and benefits of medical treatment (Leuthner 2014).

Limitations

The experiences of the parents in our study are not necessarily representative of the experiences of other parents in the same situations because we interviewed parents who contacted us. It can be speculated that parents who are willing to volunteer for empirical studies have had especially good or bad experiences. Therefore, it is possible that nonresponders differed from our participants. Consequently, the small size and qualitative design limit the generalizability of the results. Also, we did not investigate health-related data for parents (e.g., depression, anxiety, and post-traumatic stress disorder). Although their experiences were traumatic and not easily forgotten, parents recalled events that occurred in the past. Consequently, the retrospective nature of this study could have led parents to respond with socially desirable answers or to remember some experiences inaccurately. Even though we think that the richness and thickness of our data outweighed the small sample size, our explanatory model is hypothetical in nature and needs to be critically assessed in further studies with prospective ethnographic designs or parents from diverse cultural backgrounds whom we could not reach with our sampling strategy.

Implications for Care

Analyses of our findings provide specific examples of how HCPs can support parents. First, it is essential to be sensitive to the needs of parents in order to give them individual support during the EoL decision-making process. From a family-centred perspective, nurses in particular can provide parents the emotional support to create an environment in which they are involved in the care of their infants. In such an environment parents can, if they wish, take part in the decision-making process (Craig, Glick et al. 2015). A well-established continuation and coordination of care not only alleviates stress at the NICU bedside, but also can foster a trusting relationship between parents and the primary physician and nurse.

More, importantly our results show how treatment decisions are inherently connected to and reliant on the communication process. Hence, a shared and collaborative decision-making process relies on the communication skills, temporal resources, and ability to balance ethical values of HCPs (Daboval and Shidler 2014). Despite parents' wish to be involved in decision-making and HCPs increasing commitment to the topic, our findings suggest that an implementation gap regarding shared decision-making remains. It is of paramount importance that hospitals adapt their policies to fit with the current ethical standards of shared decision-making. Our results indicate that inherent problems of communication ultimately hamper shared decision-making. To prevent this, the HCPs who are closest to parents should enable and encourage parents' relationships with their newborns. They should create a space in which parent preferences for passive or active roles in decision-making can be assessed and considered a shared commitment. Open and honest communication strategies, such as sharing weighted information, discussing a range of treatment options, providing parents with time to think, and building trusting relationships through the continuation of care, can enable parents to participate in decision-making based on their preferred levels of involvement.

Conclusion

The (non)involvement of parents in the EoL decision-making of their infants showed that decision-making preferences are not homogenous but are greatly case- and context- dependent. This suggests that parental preferences with regard to the degree of control over a medical decision can range from active to passive engagement. We aimed to obtain a detailed qualitative description of the EoL decision-making process experienced by parents whose newborns died in the delivery room or the NICU. Our results suggest the necessity of careful professional evaluation of parents' wishes about involvement and clear descriptions of medical facts and treatment options. Without genuine dialogue and the competence in ethical decision-making, paternalistic decision trajectories may be reproduced.

Conflicts of Interest

The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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PART III

THRESHOLD POLICIES

Fragile Lives with Fragile Rights: Justice for Babies Born at the Limit of Viability

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Author Contributions

All authors have contributed to the different parts of the manuscript, and each author has participated sufficiently in the work to take public responsibility for appropriate portions of the content. All authors have read and approved the submission of the manuscript and take full responsibility. Miss Hendriks and Dr Lantos wrote the manuscript, critically reviewed and revised the manuscript and approved the final manuscript as submitted.

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Abstract

There is an inconsistency in the ways that doctors make clinical decisions regarding the treatment of babies born extremely prematurely. Many experts now recommend that clinical decisions about the treatment of such babies be individualized and consider many different factors. Nevertheless, many policies and practices throughout Europe and North America still appear to base decisions on gestational age alone or on gestational age as the primary factor that determines whether doctors recommend or even offer life-sustaining neonatal intensive care treatment. These policies are well intentioned. They aim to guide doctors and parents to make decisions that are best for the baby. That is an ethically appropriate goal. But in relying so heavily on gestational age, such policies may actually do the babies a disservice by denying some babies treatment that might be beneficial. In this paper, we argue that such policies are unjust to premature babies and ought to be abolished. In their place, we propose individualized treatment decisions for premature babies. This would treat premature babies as we treat all other patients, with clinical decisions based on an individualized estimation of likelihood that treatment would be beneficial.

Keywords: neonatology, gestational age policies, decision-making, justice

Introduction

Decisions about whether to try to save a baby born at the borderline of viability are among the most complex decisions in clinical medicine. The parents are stressed, the prognosis is uncertain, the treatment is expensive and burdensome, and the consequences of being wrong can be tragic. Given these complexities, it is not surprising that doctors and policymakers have often proposed guidelines for making such decisions or that those guidelines rely heavily on the estimated gestational age (EGA) of the baby. Such guidelines reduce the complexity and allow decisions to be made quickly and without the uncertainty that would inevitably accompany a more individualized treatment approach. The problem with such guidelines is that a large body of evidence suggests that EGA alone is inadequate to accurate prognosticate the likely outcomes (Tyson, Parikh et al. 2008). In spite of this evidence, many professional societies and many national health systems throughout Europe and North America still have policies regarding the treatment (or non-treatment) of premature babies, and many of those policies rely heavily on EGA. Guillen and colleagues recently reviewed 34 different policies from 23 countries and 4 international groups. Most recommended only comfort care for babies born at 22 weeks. Not a single one recommended routine or universal active care for babies born at 22 or 23 weeks (Guillen, Weiss et al. 2015). Although some efforts have been made to establish an individualized care approach, current problems still exist at the implementation level. This article will address the ethical problems with such policies and suggest a preferable moral framework for future policies.

Such EGA-focused policies reveal an inconsistency in the approach to preemies compared to other patients. In other high-risk clinical situations (e.g. patients with cancer, stroke, or major trauma), doctors are generally encouraged to consider all of the factors that make each individual patient's situation unique (Jauch, Saver et al. 2013, American Heart Association and American Stroke Association 2016). For these other patients, the indications and contraindications for treatment are all considered relative, not absolute, and doctors, patients, and family members are expected to deal with the irreducible complexity. Doctors try to make an individualized assessment of the patient's prognosis and, if there is a reasonable chance of a good outcome, they recommend treatment (Extracorporeal Life Support Organization (ELSO) 2013, Montori, Brito et al. 2013). If doctors think that the outcome will be bad, they discuss that with the patient or surrogate and may recommend palliative care only (Bossaert, Perkins et al. 2015). When there is a reasonable chance of a good outcome, they might try to guide surrogates towards one decision or another by either discouraging or encouraging treatment.

There are many factors that might alter the prognosis for a tiny premature baby. These factors include, among others, birth weight, gender, race, and whether antenatal steroids were given to the pregnant woman. These factors can dramatically alter a baby's chance of survival, so that, for example, a black female baby whose mother received steroids has a much higher likelihood of survival that a white male whose mother did not receive steroids, even at the same gestational age (Tyson, Parikh et al. 2008). Nevertheless, in many centres, the 23-week girl with a better prognosis would be denied treatment that is provided to the 25-week boy who is likely to do worse (Guillen, Weiss et al. 2015). These modifying factors have led some professional societies to recommend individualized decisions that do not rely so heavily on EGA. But, as noted above, many policies continue to focus on EGA and there is evidence that many doctors in many centres do not individualize decisions for such babies (Marmion 2017, Ruthford, Ruthford et al. 2017).

This is an exceptional situation in clinical medicine. Generally speaking, when treatments are available that offer a 20-60% chance of survival, doctors are eager to offer such treatments (Tomlinson and Brody 1990, Iserson 1996). But, for premature babies, oddly, some centres continue to not even offer treatment (Berger, Bernet et al. 2011, Geurtzen, Draaisma et al. 2016, Perlbarg, Ancel et al. 2016). Not surprisingly, centres that do actively treat these babies report higher survival rates than in those centres where premature babies are not offered treatment (Fellman, Hellstrom-Westas et al. 2009, Ishii, Kono et al. 2013, Rysavy, Li et al. 2015, Mehler, Oberthuer et al. 2016).

Janvier and colleagues have offered an explanation for this unique inconsistency in medical practice. They have shown that most people think about treatment decisions for preemies differently than they think about similar decisions for older patients. Moreover, they are less willing to provide medical care to extremely premature babies than to other patients, even when the baby has a better prognosis than the other patients (Janvier, Lantos et al. 2008, Dupont-Thibodeau, Hindie et al. 2017). This is true for both doctors and parents, both of whom seem to think of and treat the premature baby as a 'conditional person' (Forman 2009, Fei 2015). These attitudes undergird the EGA-focused policies that lead to non-treatment of many premature babies.

In this paper, we will suggest that non-treatment of babies who have a reasonable chance of intact survival is contrary to both medical norms and to most theories of justice. But before delving into these arguments, we begin with some facts about the realities of neonatal intensive care today regarding survival outcomes and practice variation. Then, we will present the reader with six

rationales behind EGA-based treatment policies and argue that these rationales do not withstand critical scrutiny.

NICU Care Today

Neonatologists consider GA by weeks and days. When we (or treatment policies) discuss babies born at 24 weeks of gestation, this refers to babies born between 24 weeks and 0 days (24 0/7) and 24 weeks and 6 days (24 6/7). This article focuses on premature births at 22 0/7 weeks to 24 6/7 weeks of gestation. We refer to babies born at <25 weeks of gestation as extremely premature babies (EPBs).

Of note, although policies and clinical studies discuss gestational age as if it can be determined precisely, in most cases it cannot. GA is determined prenatally by two factors, the mother's recollection of her last menstrual period and an ultrasound examination of the foetus. The exception is in cases of in vitro fertilization, in which the day of conception is known precisely. Consequently, unless a women and/or family has opted for assisted reproductive technology, GA cannot be known with certainty. Prenatal assessment of GA (by last menstruation period, clinical assessment, and/or ultrasound) can be wrong by as much as a week or two in either direction (Spong 2013, Butt, Lim et al. 2014). After the baby is born, doctors estimate GA by certain features of the physical examination. These, too, have a margin of error of a week or two (Dietz, England et al. 2007, Wingate, Alexander et al. 2007). Thus, when we say that a baby is born at, say, 23 weeks, we really mean that the baby is somewhere between 21 and 25 weeks of GA. Hence, most discussions accurately use the term 'estimated gestational age.'

Additionally, current outcomes are determined by the conjunction of two major factors: by a) the efficacy of treatment itself; and (b) the policies that permit or prohibit treatment of particular groups of babies. These two points will be discussed in the following paragraphs.

Outcomes for Babies Born at the Borderline of Viability

Advances in perinatal and neonatal care have significantly improved the survival of babies born at <25 weeks of gestation (Mehler, Grimme et al. 2012, Ishii, Kono et al. 2013, Rysavy, Li et al. 2015, Mehler, Oberthuer et al. 2016). Survival rates remain higher for babies born at greater gestational ages. Some centres have observed not only a decrease in risk of death, but also a decrease in long-term neurodevelopmental impairment among survivors, even for babies born at 22 to 24 weeks of gestation (Younge, Smith et al. 2016).

In spite of this potential for good outcomes, many professional societies and NICU centres throughout Europe and North America discourage or do not offer treatment to any babies born at <24 weeks of gestation (Neonatal Subcommittee of the Irish Faculty of Paediatrics 2006, Verloove-Vanhorick 2006, De Laat, Wiegerinck et al. 2010, Moriette, Rameix et al. 2010, Rutkowska 2011, Mendes and da Silva 2012, Guillen, Weiss et al. 2015, Kaempf, Tomlinson et al. 2016, Berger, Steurer et al. 2017). One justification for EGA-focused policies is that they are based on local outcome data that differ from what is reported in the peer-reviewed literature. Some doctors claim, for example, that, although some specialized centres achieve high survival rates for babies at these gestational ages, their own centre does not. These policies, then, are justified by a claim that they reflect actual survival rates at the centre (or country) in which they are promulgated. Such arguments result in self-fulfilling prophecies. If no babies are treated at 22 or 23 weeks, then no babies will survive (Mercurio 2005). To make an analogy, this would be as if some centres treated leukaemia and reported 50% survival rates when other centres did not offer treatment and reported 100% mortality. It would be illogical for the non-treating centres to use their 100% mortality to argue that treatment was futile and should not be offered. Evidence suggests that any tertiary care centre that makes a commitment to treat tiny babies will see improved survival rates (Serenius, Sjors et al. 2014).

Different policies and practices make it difficult to interpret divergent data on neonatal survival outcomes. To interpret the data, we would need to know whether there were differences in obstetric practices (e.g. induction and caesarean section) but these are often not reported (Marlow 2015, Rysavy, Li et al. 2015). We also need more detailed data than is often provided about the exact cause of babies' deaths. Often studies do not distinguish between, for example, a baby dying *with* a parenchymal haemorrhage for whom life-support was withdrawn and a baby dying *because* of a parenchymal haemorrhage (Verhagen and Janvier 2016). The former might be an elective withdrawal of treatment based on considerations of quality of life, while the latter might reflect the futility of attempted treatment. Some studies do not differentiate between birth outcomes after active treatment and withholding treatment (Smith, Blondel et al. 2017). This can be misleading when used to infer the probability of a good outcome for 22-weekers in which treatment is intended. Studies show outcome results that are the average survival rates of all babies among centres with different philosophies (Ishii, Kono et al. 2013). But this is as irrelevant as reporting the survival

rates from cancer which includes patients who chose chemotherapy as well as patients who chose to forego life-sustaining treatment.

To address these inconsistencies, a group of scholars have recently proposed standards for outcome studies in neonatology that would include data on decisions regarding treatment. They note that results on survival can only be interpreted accurately if we know the frequency of decisions to provide or withhold life-sustaining treatment since these decisions powerfully influence reported survival statistics (Rysavy, Marlow et al. 2016). Until such reporting practices are followed, published studies must be interpreted with caution born of knowledge of large variations in practices.

Evidence of Practice Variation

There is clear evidence of practice variation among different centres in both Europe and North America. In Europe, there is little consensus regarding the treatment of babies less than 25 weeks EGA. In Germany, most babies born at 22 and 23 weeks receive active treatment. German centres report higher rates of survival at these gestational ages than most other countries (Mehler, Oberthuer et al. 2016). Reports from some NICUs in Sweden show high survival rates for babies born at 23 weeks who received active interventions (Serenius, Ewald et al. 2016). Other Swedish centres are less likely to treat babies born at this gestational age and have lower survival rates. Regional differences in mortality rates were predominantly influenced by local policy practices for delivery and management immediately after birth (Serenius, Sjors et al. 2014, Serenius, Blennow et al. 2015). In many European countries, national policies recommend no interventions for babies born at 22 or 23 weeks (Gallagher, Martin et al. 2014, Guillen, Weiss et al. 2015). In France and the Netherlands, there is usually no active intervention at <24 weeks. Not surprisingly, mortality for babies born at 22 and 23 weeks in these countries is >90% (Geurtzen, Draaisma et al. 2016, Perlbarg, Ancel et al. 2016), much higher than reported mortality rates for such babies in Scandinavian countries or in North America where treatment is provided (Fellman, Hellstrom-Westas et al. 2009, Rysavy, Li et al. 2015).

In North America, there is tremendous variation in the treatment of babies born at 22 and 23 weeks of gestation. Rysavy et al showed that, among 24 academic medical centres in the US, seven offered ‘active treatment’ to all babies born at 22 weeks, four did not offer such treatment to any babies born at this EGA, and 13 offered treatment to some but not others. If an individualized approach was being used, we would expect that some babies born at 22 weeks would receive

treatment whereas others would not. This seems to be practice at some hospitals, but the fact that other hospitals have a rate of active treatment of 0% or 100% suggests that other policies are at play. For babies born at 23 weeks, the same study showed that nine centres offered active treatment to almost all babies, but the rates of active treatment at the other 15 centres ranged from 25% to 85%. Instead at 24 weeks of gestation, practice variation disappeared almost entirely, with rates of active treatment >90% at all centres (See supplementary Material of Figure S1 inRysavy, Li et al. 2015). This suggests that many NICUs in the US continue to have policies based on EGA that do not resemble the individualized approach as suggested by ACOG and AAP (AAP 2009, ACOG 2015). Similarly, Hellmann et al. reported marked variation between Canadian centres regarding the 'ethical culture' of units with regard to offering or withholding life sustaining measures (Hellmann, Knighton et al. 2016).

These data also suggest ways in which 'the language of individualized care' can be misleading. Some treatment decisions are individualized for some babies at some centres, but, at many centres, decisions are driven more by EGA-based policies than by individualized decisions. There is wide variation regarding the gestational age at which it is considered appropriate to encourage individualized care. Currently few policies recommend individualized care for all babies born between 22 and 25 weeks of gestation (AWMF 2014, ACOG 2015, Guillen, Weiss et al. 2015). Often, an individualized approach is suggested within the grey zone. Policies from the UK and Sweden consider an individual approach appropriate at 23-24 weeks, and Canada at 23-25 weeks (AWMF 2014, ACOG 2015, Guillen, Weiss et al. 2015). These statements go against their own claims regarding the limitations of gestational age thresholds. Why do some policies set the limit of an individualized approach at 23 weeks of gestation, if we know that a 22-weeker can have a better predicted outcome than a 23-weeker?

The promulgation and use of policies that rely primarily on gestational age seems to be unscientific, inexplicable, and unjust. Why, then, do such policies persist? We now delve into the six rationales for EGA-focused policies.

Rationales for Treatment Policies Based on EGA

The continued use of policies to allocate treatment based on EGA is buttressed by six different sets of considerations. One focuses on the burden of treatment. The second focuses on.EGA as an

accurate predictor of long-term neurocognitive impairments. The third focuses on the continuum between prenatal and postnatal decisions. A fourth focuses on questions of whether a newborn premature baby is a person with human rights. The fifth focuses on the relative burden of death. The sixth focuses on costs.

The Burden of Treatment

One of the distinctive features of intensive care for extremely premature infants is that the burden of treatment is substantial (Wilkinson 2012). In general, reductions in gestational age are associated both with worse prognosis and with greater burden of treatment (Stoll, Hansen et al. 2015). This double jeopardy of worse prognosis and increased burden can be one reason why EGA may have a closer relationship to withholding treatment than other prognostic factors. It is also one potential reason why treatment is withheld from extremely premature babies when it would be provided to other patients with similar prognoses.

Although it is true that projections in gestational age are associated with infants' need of longer periods of support and higher risk of various complications, it is only incrementally higher in each week of gestational age. Serenius et al. have shown that neither gestational age nor birthweight was a significant determinant of major short-term morbidity (e.g. necrotizing enterocolitis, severe chronic lung disease, retinopathy, etc.) among survivors born at 23-25 weeks. No obvious demarcation line exists in which morbidity drastically increases among survivors born at 23 or 25 weeks (Serenius, Ewald et al. 2004). Further studies are necessary to investigate these short-term outcomes by week of gestational age. Although the burden of treatment can outweigh the overall benefits of treatment, this is not the case for every EPB born <25 weeks gestation. Instead, physicians' ought to disclose these burdens to parents and engage them in shared decision-making, rather than following a 'one-size-fits-all' policy.

EGA as Predictor of Long-Term Neurocognitive Impairment

It is well known that extreme prematurity is associated with brain damage and long-term neurocognitive impairment. This data comes from studies comparing premature babies with term babies. Preemies clearly and consistently have higher rates of seizures, cerebral palsy, learning disabilities, and autism (Jarjour 2015, Stoll, Hansen et al. 2015).

The data are less clear when comparisons are not between preemies and term babies but between babies born at different gestational ages between 22 and 25 weeks. The relevant

comparison for policies based on EGA is the comparison between babies born at 22-23 weeks and babies born at 24-25 weeks. That, after all, is the comparison that would inform policies based on differential treatment for babies born at these gestational ages.

Although it is also well-known that babies at 22 and 23 weeks have much lower rates of survival than babies born at 24 or 25 weeks, this, too, may not be the most important consideration. If many babies born at 23 weeks die, but most of the survivors are neurocognitively intact, then treatment might be medically indicated and ethically justifiable. Tiny babies would be analogous to adults with some cancers for whom treatment is unlikely to be successful but, if successful, would lead to long-term intact survival. In the adult situation, treatment would likely be offered (Reyna, Nelson et al. 2015). As we have seen, for preemies, it is often not.

This raises a question on the rate of neurodevelopmental impairment among survivors. If all survivors are severely impaired, one might decide that treatment should not be offered. If most survivors were neurologically intact, then treatment should be offered. Unfortunately, neither of these extremes is true. In fact, EGA is not highly correlated with long-term outcomes among the tiniest babies. Several studies show that, among survivors, neurodevelopmental outcomes are similar for babies born at 22, 23 and 24 weeks. Wood et al, in the EPICURE study, showed that, even in the late 1990s, survivors born at 23 weeks had similar neurocognitive outcomes to survivors born at 24 and 25 weeks (Wood, Marlow et al. 2000). More recently, Andrews et al. displayed that survivors' neurodevelopmental impairment was unaffected by their gestational age. Instead, they showed that EPBs who survived with or without neurodevelopmental impairment did not differ from each other in their EGA or birth weight (Andrews, Lagatta et al. 2012). An analysis of the database of the Italian Neonatal Network showed that, though lower EGA was associated with neurodevelopmental impairment, the predictive value was 'too low to be useful in predicting individual outcomes' (Gagliardi 2015). Clearly, an increase in EGA is not reliably associated with better neurodevelopmental outcome (Meadow, Lagatta et al. 2012, Moore, Lemyre et al. 2013). These studies suggest that it is inappropriate to rely on EGA to decide that babies born at 22 or 23 weeks should not be resuscitated but that babies at 24 weeks or more should.

The Continuum between Prenatal and Postnatal Decisions

A third consideration that buttresses policies based on EGA has to do with the continuum between fetal life and extra-uterine life (Ohel, Levy et al. 2009). In order to achieve the best

survival rates for preemies, some interventions must be given to the pregnant woman who goes into labour early. Preemies do better, for example, if their mothers are given steroids to hasten the development of the fetal lungs. And in some cases, a C-section may be medically indicated (although there still remains some discussion in this debate, the current recommendation is to individualize decisions on delivery. For an overview see Reddy, Zhang et al. 2012, Ecker, Kaimal et al. 2016, Humberg, Härtel et al. 2017). But the decision about whether to use steroids or how to deliver is up the woman herself. At that point, she is the only 'legal' patient, even though her choice will have a profound effect on the unborn child's chances for survival.

This continuum of care can be used to argue that, because pregnant women have the right to make decisions about their own treatment during pregnancy, even decisions that will have an impact on the unborn child's chance for survival, they should also have the right to make decisions postnatally about life-sustaining treatments for their child. We believe that the continuum between prenatal and postnatal decisions has different ethical implications.

Pregnant woman, like all competent adults, have the right to make medical treatment decisions for themselves. Hence, a pregnant woman's decisions about steroid use, fetal monitoring, a caesarean section, or the timing of delivery must be respected. To do otherwise would violate women's basic rights to personal autonomy and bodily integrity (Cummings and Mercurio 2011). As the American College of Obstetricians and Gynecologists' (ACOG) opinion shows, the obstetrician may have divided loyalties, but his or her primary loyalty must be with the pregnant patient rather than with the foetus (ACOG 2016).

Loyalty to the pregnant woman, however, does not mean that the obstetrician or neonatologist should act paternalistically on her behalf. The physician has an obligation to provide the pregnant woman with information about all available options and the risks and benefits of each treatment. In clinical practice this means that when a woman goes into labour at 22 or 23 weeks of gestation, she should be given a range of options, including ones that might expose her to some risk but would offer benefits to her unborn child. She could be given steroids to accelerate maturation of the fetal lungs, provided that her goal is to provide neonatal intensive care and life-supporting interventions for the unborn. If, instead, she decided not to pursue active intervention for her unborn, then she would likely choose against steroids or other obstetrical interventions that might lead to a better neonatal outcome. Similarly, in some situations, a C-section will improve outcomes for the unborn. But this increases risks for the mother. With regard to both steroids and C-sections, the pregnant woman may refuse treatment, resulting in a worse prognosis for her unborn (Ecker, Kaimal et al.

2016, Fanaroff and Fanaroff 2016). By the same token, she has the right to choose treatment that would result in a better prognosis for her unborn. But that option is only a realistic one if treatment would be offered.

We suspect that the full range of choices is often not offered to pregnant women who go into labour at 22 and 23 weeks (Christoffersen-Deb 2012, Staub, Baardsnes et al. 2014, Boland, Davis et al. 2016, Marmion 2017, Mercer 2017, Ruthford, Ruthford et al. 2017). This would likely be true at centres that do not offer active treatment at these gestational ages. After all, it would make no sense to intervene prenatally if no postnatal interventions were to be offered.

The result of these considerations is that it is possible for women in the same high-risk pregnancy to have two opposing prenatal discussions based on the hospital's or country's EGA-focused policy rather than on the prognosis. For example, consider a pregnant woman who is in labour at 23 weeks of gestation. In some countries or centres, she would be told that, if her unborn child were born at 23 weeks, it would not receive life-sustaining treatment. So, it would be unreasonable for her to choose antenatal steroids. But if that same woman is in another centre, doctors might recommend antenatal steroids. They might also inform the woman that the prognosis will be better if she consents to treatment. In such circumstances, many women might choose treatments for themselves that have, as their goal, a better outcome for their unborn child.

We believe that pregnant women at risk for premature birth should receive individualized antenatal counselling and that decisions should be made based on the woman's goals and preferences (Gaucher and Payot 2017).

The Human Rights of the Newborn

The balance of rights changes once the baby is born alive. At that point, the baby has rights that are independent of the mother's rights. At the moment of birth, a baby can now be cared for as an individual without negating the rights of a pregnant woman. Then, the autonomous rights of the mother should no longer supersede the rights of the newborn (Warren 1997). Human rights are inalienable fundamental rights of people on the basis of their humanity regardless of their nationality, place of residence, sex, colour, religion, language or any other status (OHCHR). They are rights to which a person is inherently entitled simply because she or he is a human being.

Some argue, however, that a newborn does not have the same rights as other individuals and they do not have equal moral status (Warren 1997). To date, there is no consensus in the

philosophical literature on when a human being acquires 'personhood' and has equal moral status as other individuals who are entitled to human rights. If preemies are considered to have 'personhood', hence entitled to human rights and corresponding moral status, then treatment decisions for prematurely newborn babies should recognize their rights and moral status independently from the mother or the family. Instead, for many scholars, personhood is contingent upon adequate cognition. The basis for these arguments is that babies do not have key cognitive capacities such as sentience and self-awareness (Tooley 1972, Kuhse and Singer 1985). By these arguments, we should treat tiny preemies (and all newborns) as conditional persons who do not yet have a justifiable moral claim to equal treatment. They would acquire personhood, and thus, rights, at some later date only if they develop the appropriate cognitive capacities.

This position conflicts with the universal human rights framework that is embodied in the *UN Convention on the Rights of the Child* (CRC). The CRC recognizes that moral status begins at birth. The convention makes no distinctions between premature babies, term babies, and older children. In addition, Article 6 Section 1 and 2 state that children's inherent right to life should be recognized to the maximum extent possible, while Article 24 makes clear that the child's interests should be of primary concern (Saugstad 2016, Saugstad and Stokkereit 2016). We agree with the human rights framework of the CRC. The moment of birth is the moment at which the newborn becomes an individual and a member of the social and political community with full *legal* rights. Any other approach would require a deeply problematic attempt to draw bright lines across the continuum of human development and human capabilities (Nussbaum 2009, Wasserman, Asch et al. 2013). Ultimately, it would lead to the dehumanization of large segments of the population who may lack some threshold of cognitive capacity, including, not just preemies but, perhaps, children with neurocognitive impairments and adults with dementia (Gosepath 2011). However, this logical implication is rarely drawn and older adults with cognitive impairment, unlike extremely premature babies, are often attributed moral status in spite of their cognitive condition (Buron 2008).

The Relative Burden of Death

Another plausible explanation for EGA-focused policies is the relative burden of death. This account holds that the interest of an extremely premature baby (and perhaps to a lesser extent a term baby) in his or her future might be less than that of an older child. According to the intuition of many people, the death of a two-year old is worse than the death of a newborn. It may be that this intuition underlies an evaluation of best interests, which yields different answers for extreme

premature babies. The benefits and harms of providing versus withholding treatment for an extremely premature baby or baby born at term are evaluated differently, yielding a different outcome threshold for decisions than in older patients (McMahan 2002, Wilkinson 2013).

We grant that *theoretically* people might have this intuition. They value a one-day-old term newborn less than a two-year-old, and they would value a preemie less than a term baby. At least in the sense that they would view a death not quite as bad. But this theoretical intuition can be contrasted with the way people actually behave. When given the choice, most mothers and their partners choose to initiate care for their baby (Staub, Baardsnes et al. 2014, French 2017). This means that whilst there might be this shared moral intuition that death is *less* bad, it does not mean it is *not bad*. It also does not mean that when given a choice most people would seek to avoid it, so that making an EGA policy based on that intuition protects and helps parents. Instead, one must ask whether, from the parents' perspective, it is better to try to save a tiny baby, even if the effort is unsuccessful, than it is to not try at all. That is exactly the sort of value question that properly ought to be answered by parents rather than by doctors or policy makers. Some parents might prefer comfort care and a peaceful death, while others may want to give their baby a chance. In such situations, the best approach would be a trial of therapy. The trial of therapy might be unsuccessful, in which case the baby would die. If successful, however, the baby might have good long-term outcomes. Many studies suggest that parents are more likely than doctors or nurses to prefer the trial of therapy (Streiner, Saigal et al. 2001, Lam, Wong et al. 2009).

The Cost of Neonatal Intensive Care

A final consideration that supports EGA-focused policies are based on the fact that the care of each tiny baby is very expensive. Saving a baby born at 24 weeks costs hundreds of thousands of dollars (Partridge, Robertson et al. 2015). The high cost of treatment is sometimes used to justify policies to limit the provision of intensive care treatment (Wilkinson and Savulescu 2014). In addition, preemies have higher lifetime care costs than do other children because they are more likely to have chronic medical problems as well as neurocognitive or developmental problems. The combination of short-term and long-term costs, for both parents and society, has been invoked as valid justifications for policies that limit life-sustaining treatment (Cavallo, Gugiatti et al. 2015).

There are good ethical reasons to consider the cost of treatment in making policies about what treatments to provide. After all, it would be both unfair and unwise to provide extraordinarily

expensive treatments to one group of patients if the costs of those treatments are collectively subsidized and the outcomes are not good. Ultimately, this would lead to fewer resources available for beneficial treatments for other patients (Wilkinson and Savulescu 2014). But there are three problems with these economic arguments as they are used in the context of extremely premature babies. The first is that, like so many arguments about preemies, they are applied selectively. Many other patient groups, including people with cancer, bad heart disease, or strokes require expensive treatments that sometimes result in lifelong chronic conditions. Like preemies, patients with these conditions have a wide variety of outcomes (Go, Mozaffarian et al. 2014). But, in these settings, the possibility of a bad outcome is used to discuss risks and benefits with patients or surrogates, not to make policies about when or whether treatment should be offered (AAP 2016, American Heart Association and American Stroke Association 2016). Of note, even in European public health settings, where decisions are made about the costs of certain treatments and the price worth paying for health interventions, costs are not cited to deny all treatments or intensive care to broad groups of patients. Such discussions rather focus on specific treatment costs and pharmaceutical drugs (King Baudouin Foundation and Belgian Advisory Committee on Bioethics 2013).

The second problem with these sorts of arguments is that they would require an objective threshold of both costs and benefits that could be applied to a wide variety of patient groups in order to decide when or whether treatment should be offered. But it is difficult to develop an objective measure that will help determine whether a life is so miserable or so costly as to be not worth living. Cost-benefit analyses rely on measures such as ‘quality adjusted life years’ or QALYs to lend an air of objectivity to such discussions. QALYs adjust the value of a year of life by a factor that takes into consideration various disabilities. This methodology is useful for many things, but it requires subjective assessments of the quality of another person’s life and studies show that such assessments rarely correlate with individuals’ assessment of their own quality of life.

Finally, even considering the likelihood of at least some bad outcomes, and even considering the cost of saving a tiny preemie, neonatal intensive care compares favourably to many other seemingly comparable treatments in is cost-effectiveness (as measure by cost per QALY or any other measure of cost-effectiveness). Economists estimate that, even for the tiniest babies born <26 weeks, NICU care costs about $8000 per quality-adjusted life year. And recent studies confirm that from a maternal-neonatal perspective resuscitation of 23-weekers is cost-effective. These costs/QALY are far less than coronary artery bypass surgery. They even compare favourably to routine Pap smears, which have been estimated to cost $17,000/QALY (Cutler, Meara et al. 2000,

Korvenranta, Linna et al. 2010, Lantos and Meadow 2011, Partridge, Robertson et al. 2015). The QALY measurements take into account neurocognitive impairment and other chronic problems and reflect the fact that survivors of NICU care and their families both report a good quality of life among NICU survivors (Payot and Barrington 2011).

There are two explanations for the perhaps surprising cost-effectiveness of such expensive treatment. First, while NICU survivors with chronic health problems certainly have increased lifetime care costs, most babies who survive do not have special health needs. After age 1, the medical care costs for premature babies are similar to the costs for babies born at term (Pignotti and Donzelli 2015). Second, most babies who die tend to die relatively quickly. Those who survive are often in the NICU for months (Stephens, Lain et al. 2016). Thus, most expenses in the NICU are directed to babies who will ultimately survive and do well (Buchh, Graham et al. 2007). In this regard, NICU care is more cost-effective than intensive care for adults, where most expenditures go to the care of patients who will either die in the hospital or die within a year of discharge (Meadow, Hall et al. 2003). In adult ICUs the best outcomes are among the patients with the shortest stays. The longer a patient is in the ICU, the higher their mortality rate. Most expenses in adult ICUs are thus directed toward patients who have the worst outcomes. As a result, they are far less cost-effective than NICUs. Put in another way, the most expensive NICU patients are also the most cost-effective, whereas the most expensive ICU patients are the least cost-effective (Hayman, Leuthner et al. 2015).

To be clear, we believe that cost-effectiveness analysis can and should be considered in developing just policies to allocate health resources. But, to be just, these policies should treat all patients equally in the sense that they should apply similar cost-effectiveness criteria across the entire population.

Differential treatment based on cost-effectiveness could be ethically justified as an appropriate allocation of resources. Prioritization of limited life-saving resources is not discriminatory or unjust when the choice to forego treatment for an infant is based on ethically relevant and consistent considerations (Wilkinson and Savulescu 2014). But such a utilitarian argument is not consistent if it is applied *only* to preemies. A justification for non-treatment of these babies that is not accompanied by a comparison of the utility of such treatment with the utility of, say, the treatment of 80 year olds, is not true to its own first premises.

Conclusion

We have argued that policies that limit life-sustaining treatment based on the estimated gestation of an extremely premature baby neither medically sound nor just.

They violate egalitarian principles because they do not provide equal access and fair chance to all individuals with a similar prognosis (Savulescu 2002). Additionally, a Rawlsian model of justice would only permit differential allocation of resources as long as that allocation preferentially favoured the most vulnerable citizens. Such an approach would seem to dictate that preemies be given preferential access to treatment, rather than face programmatic barriers to treatment. The health burdens of premature babies can be understood as an arbitrary effect of a natural lottery and bad luck. A just health care system should mitigate the unpredictability of nature through social institutions that assure equal opportunities despite biological differences and bad fortune (Daniels 2001). Moreover, a just health care system would not allow resources to be allocated preferentially to able-bodied people compared to disabled people. If anything, the opposite should occur, in order to equalize the capabilities of both groups (Nussbaum 2009).

We assert that newborns, even those born prematurely, are full-fledged members of the human community and have the same right to treatment as other humans. This entails giving extreme preterm babies the best possible chance to life. Policies and practices that limit treatment based on gestational age alone violate this basic principle of justice.

Taken together, these arguments lead to a clear conclusion. Policies that limit treatment based on EGA for babies who could survive with treatment violate important principles of justice. Instead, parents, doctors, and health systems should insure that each patient, even if the patient is a tiny preemie, be assessed as an individual. Treatment should be provided if, after such an assessment, it is likely that the treatment will be beneficial for that individual. The criteria for deciding whether a treatment is likely to be beneficial should be the same for extremely premature babies as they are for other individuals.

The criteria for limiting treatment in preemies, as in any patient group, should include a combination of (1) poor prognosis for survival, (2) likelihood of unacceptable disabilities, (3) burdens of treatment, (4) costs and cost-effectiveness, and 5) informed consent and shared decision-making with parents. A just system would apply those criteria to all patient groups. Our current approach of denying treatment to some premature babies when it is provided in situations where it is less cost-effective to patients with a worse prognosis is unjust.

This might be perceived as a radical change. But it should not be. After all, there are no national policies regarding other patient groups. We do not limit treatment of the elderly based on age alone, or of cancer patients based only on the staging of their tumour. Our proposed approach simply recognizes that gestational age is a less powerful predictor than some believe it to be and that premature babies are more unique than they are sometimes seen to be. These policies, present in European but also in North American NICUs, could be an expression of a culture that systematically denies care for babies of certain gestational ages. It is important to call out on policy-makers and their responsibility in rejecting EGA-focused policies. Changing policies is but one step in a long process of cultural change. Consequently, it is necessary to continue to address and discuss the limits of EGA within the neonatal community to foster a cultural change in the way physicians treat babies born extremely premature.

We are not suggesting that all premature babies should be treated. We are making a more modest recommendation. We believe that all premature babies should have access to diagnostic and therapeutic regimens that are comparable to those that are available to other patients with life-threatening illnesses. There will be situations in which, after a thorough assessment, doctors and parents decide that the potential burdens of treatment outweigh the potential benefits. There will be situations where treatment is deemed futile. There are many situations in which a choice for palliative care rather than intensive care is appropriate and ethically justifiable. But we do not believe that this is true for all babies born at the borderline of viability, and it is especially not true for babies born at an EGA of ≥22 weeks. With regard to such babies, treatment decisions should be made after an individualized assessment of prognosis for survival and considerations of likely impairments. Such an individualized approach recognizes that simple rules are unlikely to solve complex problems (Dupont-Thibodeau, Barrington et al. 2014).

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GENERAL DISCUSSION

Main Findings

This final chapter integrates the outcomes of each individual study and discusses their findings in a broader context. In this section, I will proceed as follows. First, I will summarize the main research objectives and present the overall findings. Then, I will put the findings into a larger perspective and I will critically review the methodology of the three studies. I will end this chapter presenting some implications for practice and future research with some suggestions to improve the process of end-of-life decision-making for infants born extremely premature.

Part I – Societal Attitudes and Values

Box 3. Objectives of the Study

- To provide insight into the effect of societal attitudes and values on end-of-life decision-making for extreme preterm infants;
- To explore whether socio-cultural differences would lead to diverging end-of-life decisions;
- To identify the willingness to carry the costs for extremely preterm infants at risk for disabilities in different linguistic parts of Switzerland.

The main finding of the population survey is that the majority of the Swiss population preferred a shared decision-making model in which parents and health care professionals make shared decisions for LSIs of infants born extremely premature.

Furthermore, the context of socio-cultural demographics, especially the linguistic region, influenced attitudes towards extreme prematurity and decision-making. For example, people from the Italian-speaking region were more in favour of giving physicians sole authority to make decisions. In cases of disagreement between neonatal HCPs and parents, the German-speaking population was more likely to regard the parents as the final decision-makers, while the French- and Italian-speaking populations felt that the physician should have more decisional authority. These diverging choices may be caused by Switzerland's distinct cultural composition. Another insight from the population survey is that different criteria's and values – e.g. quality of life in terms of independence or normalcy – might influence decision-making processes in daily practice. For example, lay people understood quality of life in terms of independence and/or normalcy.

Taking into account these different values can help HCPs address parental concerns more appropriately; i.e. considering not only the physical well-being of infants but also the potential for independence in the future (Hendriks, Klein et al. 2017). Furthermore, the majority of the Swiss population rated their own solidarity toward disabled persons as high. Interestingly, the Swiss population does not relate solidarity to merely financial costs. These attitudes and values have the potential to affect the level of support for parents. This may alleviate some pressure on parents and health care providers in the decision-making process of extreme preterm infants (Hendriks, Bucher et al. 2017).

Part II – Parental Experiences

Box 4. Objectives of the Study

- To define parents' perceptions toward end-of-life decision-making, communication, information and care in this process;
- To explore parents' experience of the dying process of their child at the perinatal or neonatal stage;
- To identify parents' experience of parenthood and support they found particularly helpful or challenging.

The parental interview study has shown how experiences of parents are not homogenous but highly case- and context-dependent. Unclear or inadequate communication and information about treatment options in the decision-making process was one of the most important factors causing friction and distress between parents and health care staff. At the time of the interview, some parents in our study reported that they felt uneasy about not having been adequately informed about prognostic uncertainties and about the various treatment options available at the prenatal stage. In two cases, parents commented on the hospital's internal policy on limiting treatment of premature babies at a certain gestational age. They expressed the need of an explanation of the ethical and medical arguments for withholding life-sustaining treatment and/or resuscitation. All parents who participated in our study expressed the wish for transparent information. In the NICU, a minority of parents recalled active involvement in decision-making, while most parents in our sample were not aware of being involved in decision-making. Most parents did not think that there was anything to decide, respectively that they had to make a decision themselves. The situation was evident and

'clear', the physicians' evaluation and guidance were perceived as reasonable, or it was perceived that the child had already made the decision him- or herself. This might have been influenced by the ethical consultations used at the time of the infant's admittance in the NICU. The interviewed HCPs in our study confirmed that the health care team would discuss at least three different treatment options and then deliberate until reaching a team consensus on the best treatment option. The health care team would present this proposition of care to the family, who would be asked to consider it with a right to veto. This finding raises the concern that some decisions may not have been fully shared between parents and HCPs (Hendriks and Abraham 2017).

The parental experiences of an extreme premature birth and death has shown that the parents under study experienced a multitude of stressors at various stages in which medical uncertainties played a central role. Even though parents are assigned parenthood at birth, often they cannot act as such (embodied parenthood). For those parents, whose infant was transferred to the NICU, parenthood was challenged by the separation from their child immediately after birth, by the alienating setting of the NICU, by the physical distance to the child and the inability of parents to take care of their child. It was only when the infants' situation deteriorated and death was approaching that child-parent bonding and memory making could occur. During the period after the death of the child, some parents, and especially mothers, experienced feelings of distress, shame, failure, guilt, and helplessness. The burdensome event of an extreme premature birth and subsequent death highlighted the need in professionals support to help transform from assigned parenthood to embodied parenthood (Abraham and Hendriks 2017).

Part III – Threshold Policies

Box 5. Objectives of the Study

- To explore the reasons behind policies that use the infants' gestational age as the primary factor to determine whether to recommend life-sustaining interventions;
- To reflect on the ethical permissibility of such reasons.

Based on the empirical findings of the parental interview study, the question can be raised whether parents' recollections described situations where decisions regarding LSIs for babies at the limit of viability were influenced or guided by the hospital (or national) policy of gestational-

based thresholds for interventions. Of note, this is a speculation that comes with limitations, which are further discussed in the next paragraphs. Therefore, in this third study, we analysed the literature – mostly from North America and Northern Europe – regarding ethical justifications of policies that are based on gestational age alone or on gestational age as the primary factor in the determination whether HCPs recommend or offer LSIs. In doing so, six different sets of justifications were analysed and/or disputed, i.e. the burden of treatment, the estimated GA as an accurate predictor of long-term neurocognitive impairments, the continuum between prenatal and postnatal decisions, human rights, the relative burden of death, and the costs of neonatal intensive care.

In relying so heavily on gestational age, such policies may actually do babies a disservice by denying some babies treatment that might be beneficial. Instead, policies should move away from categories of gestational age and encourage doctors to assess each baby clinically and make an individualized treatment decision, just as they do for every other patient. The criteria used to make such a decision will have to be a combination of (1) poor prognosis for survival, (2) likelihood of unacceptable disabilities, (3) burden of treatment, and (4) the overall costs of care for this patient group. If those criteria are applied to all patient groups – and not just preterm babies – it could be considered just. If other patients with a worse prognosis and higher costs or lower cost-effectiveness are offered treatment, this should be considered unjust (Hendriks and Lantos 2017).

A Broader Perspective on the Findings

End-of-life decision-making has become the subject of much research within neonatology, obstetrics and ethics. This is illustrated by the large amount of articles I referred to in the introduction of this thesis, a list that is far from complete. In reviewing the studies published in the last decades, I would like to point out three patterns that relate to my main findings.

Attitudes and Values

Divergences in attitudes and values on end-of-life decision-making for extreme preterm infants are not an unexpected result and has been shown in numerous studies that report on the attitudes and values of lay people, parents and HCPs (Cuttini, Kaminski et al. 1997, Norup 1998, Støre Brinchmann, Førde et al. 2002).

Many studies on lay peoples' attitudes and values have shown wide differences in opinions regarding limiting treatment and EoL decisions (Norup 1998, Teisseyre, dos Reis et al. 2009, Teisseyre, Vanraet et al. 2010). Our population survey has shown that the attitudes and values were divergent and reflected differences depending on the socio-cultural factors (i.e. linguistic region and age). Our findings corroborate that cultural and societal factors affect practice regarding end-of-life decisions of extreme preterm infants (Rebagliato, Cuttini et al. 2000). Furthermore, a majority of lay people in our study agreed that shared decisions between HCPs and parents was the best decision-making model. This is in line with the results from other studies on the importance of considering parental wishes and attitudes in decisions on life-sustaining treatment (Norup 1998, Teisseyre, dos Reis et al. 2009, Teisseyre, Vanraet et al. 2010).

Studies on parents of extreme preterm infants depicted a variety of experiences and attitudes toward parental participation (Lee, Penner et al. 1991, Griessel, Joubert et al. 2012). Some studies described how parents wish to be involved to some extent without being burdened by a decision (Støre Brinchmann, Førde et al. 2002, Caeymaex, Speranza et al. 2011), while other studies showed that parents want to have a more active role in the decision-making (De Vos, Seeber et al. 2014, de Vos, Bos et al. 2015, Weiss, Barg et al. 2016). Our parental interviews indicated that decision-making preferences are not homogenous but are highly case- and context- dependent. This suggests that parental preferences with regard to the degree of control over a medical decision can range from active to passive engagement.

Although beyond the scope of this thesis project, the attitudes and values of HCPs are important to take into consideration when discussing the overall impact of attitudes and values on decision-making. The majority of studies focus on the HCPs attitudes, since these are likely to impact on the presentation of information and discussion of options with parents (Rebagliato, Cuttini et al. 2000, Cuttini, Casotto et al. 2009, Garel, Caeymaex et al. 2011, Gallagher, Marlow et al. 2012, Mills, Janvier et al. 2015, Lee, Cho et al. 2017). Studies have reported that HCPs 'value their own considerations over parental wishes' as being the 'best' answer in the quest for the best interest of infants (Peerzada, Schollin et al. 2006, Haward, Kirshenbaum et al. 2011, Doucette, Lemyre et al. 2017). But the opposite also exists. HCPs are reported to follow family's wishes of withholding or withdrawing treatment even though they considered this was not in the best interest of the infant (Moratti 2010, Dupont-Thibodeau, Hindie et al. 2017). Hence, there is wide variation in attitudes and the cultural background seems to shape the health care professionals' attitudes

toward parental participation and proactive (also known as active, liberal) or restrictive (a.k.a. conservative) management (Cuttini, Rebagliato et al. 1999, Peerzada, Richardson et al. 2004, Janvier, Lantos et al. 2008).

Shared Decision-Making

Our parental interview study found that some parents did not recollect being involved in EoL decisions at the limit of viability and, in some cases, after admission of the infant to the NICU. Although it is important not to discredit parents' recollections of the decision-making process, it is important to realize that sometimes there is a difference between what parents are told by HCPs and what parents hear and understand. Consequently, what parents reported in their recollections of the events may not be a one-to-one translation of the actual discussions with the health care team (Zupancic, Kirpalani et al. 2002, Kett, Mohamed et al. 2016). This discrepancy is shown in the study of Keenan et al. (2005) in their comparison of mothers' and health care professionals' perceptions of counselling. Although HCPs reported they discussed several treatment options regarding resuscitation measures, a majority of mothers believed that they were not offered a choice (Keenan, Doron et al. 2005). Similarly, Boss et al. (2008) showed that few parents recalled discussing resuscitation options even when these discussions where documented in the medical chart (Boss, Hutton et al. 2008). Hence, it might be the case that the parents in our study did not recollect all discussions with the health care team, possibly due to the emotional and time-intensive period they went through or to better cope with the situation and the EoL process in the long term (Hendriks and Abraham 2017). For these reasons, it is important to place the parental experiences in a broader context and assess whether the experiences of the parents in our study are corroborated by other studies on decision-making and communication with parents and HCPs.

Several studies show that parents wish to be informed and actively involved in resuscitation decisions for their infants at the limit of viability (Janvier, Barrington et al. 2014, Staub, Baardsnes et al. 2014, Haward, Gaucher et al. 2017, Ruthford, Ruthford et al. 2017). This is similar for decision-making in the NICU. A nationwide questionnaire in Switzerland on parental EoL experiences found that decisions in neonatology were significantly less often shared between parents and the health care team than EoL decision-making for children in oncology, neurology or cardiology (Zimmermann, Bergstraesser et al. 2016). Furthermore, the parents in our study reported diverse preferences regarding their involvement in the decision-making. Some parents who were not actively involved wanted to be, whereas others explained they trusted their HCPs in making

the decisions. Such diverse preferences to participate in life-support decisions about their infants' care have previously been reported (Moro, Kavanaugh et al. 2011, Weiss, Barg et al. 2016). Parental engagement in NICUs has shown to be influenced by cultural and socioeconomic differences and parental readiness to be involved in the care of their infant will vary by parent ability, health status of the baby, time and resources (Benzies 2016). Both empirical studies as well as ethical analyses have suggested that parental involvement should be tailored to individual preferences and needs (Racine and Shevell 2009, Madrigal, Carroll et al. 2012, De Vos 2015, de Vos, Bos et al. 2015).

As opposed to sharing decisions through exchanging information between HCPs and parents, deliberating about all treatment possibilities, and making a joint decision about which treatment should be implemented (Charles, Gafni et al. 1999, p. 654, Payot, Gendron et al. 2007, Légaré and Witteman 2013), the findings in our parental study seem to point towards a difficulty in 'fully' sharing decisions. As stated by the interviewed HCPs in our study, parents were often recommended and asked to consider one treatment option that was considered to be in the best interest of the infant by the health care team. This is consistent with reports from other studies within neonatology (Keenan, Doron et al. 2005, Payot, Gendron et al. 2007, De Vos, Seeber et al. 2014). Then, parental choice is situated in the ability to accept or reject (i.e. veto) the proposed course of action (Keenan, Doron et al. 2005, Payot, Gendron et al. 2007). This takes the decision away from parents and can lead to situations where parents' preferences are taken only into account when there is a disagreement and further reassessment of a decision. Consequently, this enables parents with strong objections to potentially influence the decision-making process, more so than parents who comply with the proposed decision (De Vos, van der Heide et al. 2011, De Vos, Seeber et al. 2014). These findings can be understood in light of the previously mentioned ethical decision-making model in the University Hospital at the time of the events (Arlettaz, Mieth et al. 2005, Baumann-Holzle, Maffezzoni et al. 2005). However, this ethical decision-making model was in transition toward a shared decision-making model since 2014 until being officialised at the end of 2015 (Fachgruppe Ethik Neonatologie USZ 2017). It is, therefore, possible that within this transition phase HCPs were applying different ethical consultations. This transition might have differently affected cases and could have impacted how parents were involved in the decision-making process and, as a result, their recollections of the events.

High Quality Communication

As pointed out, communication can be particularly difficult since what parents hear and understand is not necessarily the same (Zupancic, Kirpalani et al. 2002, Keenan, Doron et al. 2005, Boss, Hutton et al. 2008, Kett, Mohamed et al. 2016). Numerous studies have explored how to communicate in the best way in these difficult and highly sensitive situations. Some elements have shown to prove helpful, although when not appropriately applied, they could challenge communication instead. Our study confirmed that a high quality of communication between parents and the health care team is crucial in the context of an extremely premature birth.

Good communication in the manner of providing accurate and transparent information can support parents in manifold of ways. In our study, transparent and honest information enhanced communication between parents and the health care team and helped building a trusting relationship with HCPs. According to studies on parental experiences with counselling, transparent information can help parents to cope with the situation and feelings of shock and uncertainty. Additionally, parents feel more in control and are less fearful when provided accurate information (Kavanaugh 1997, Sharman, Meert et al. 2005, De Rouck and Leys 2009, Branchett and Stretton 2012). Carefully and clearly formulated information can also aid parents in decisions and can reduce feelings of anger and distress (Davies, Contro et al. 2010). Studies have shown that these elements contribute to decision-making and parental well-being (Brosig, Pierucci et al. 2007, Obeidat, Bond et al. 2009, Xafis, Wilkinson et al. 2015, Zimmermann, Bergstraesser et al. 2016).

However, it is crucial that communication is delivered in a timely and sensitive manner (Kavanaugh 1997, Sharman, Meert et al. 2005, Branchett and Stretton 2012). In fact, good communication goes further than *what* is said, it is also about *how* a message is communicated non-verbally (Wigert, Dellenmark Blom et al. 2014). This has also been shown in our study where parents reported how empathy, familiarity, and HCPs simply ‘being there’ for parents made them feel taken seriously and supported. Henley & Schott (2008) emphasize the importance of sensitive language when approaching parents, especially parents who just lost their child. This means avoiding clinical terms, and instead talking about the baby in a person-like manner respecting his or her gender and first name. This also relates to our findings on parenthood in the NICU that involves viewing and treating the baby as a person, and parents and the baby as a family, even after the baby has died. Such non-verbal communication is part of providing information sensitively alongside avoiding to provide parents with too much information (Boss, Donohue et al. 2016, Ruthford, Ruthford et al. 2017).

Another example that can challenge or establish good communication between parents and HCPs is continuity of care. Neonatal intensive care is provided by many professionals with different sets of expertise. Studies show that consistency and continuity in sharing information should be maximized for parental satisfaction (Orfali and Gordon 2004, Hellmann 2014, Wigert, Dellenmark Blom et al. 2014, Kenner, Press et al. 2015). Some parents in our study experienced challenges related to fragmentation of care, but not as much within hospital wards as between hospital and outpatient care. After the child's death, mourning parents received (mainly) written information on supportive outpatient services such as postnatal exercise groups for parents of deceased infants or psychologists. However, the organization of the support of these services were often left to the parents. This shows how the appropriate care structures were in place for these parents, following the principles from neonatal palliative care as described by Stafford (2015) and Ahern (2013), but were fragmented in their utilization. Ideally, in neonatal palliative care, psycho-social and bereavement assistance to parents is not limited to the hospital stay but is initiated—in the case of anticipated extreme prematurity—before hospital admission and continued after hospital discharge and, thus, does not end with the child's death (Leuthner and Jones 2007, Williams, Munson et al. 2008).

Hence, transparent communication, non-verbal communication and continuity of care are among the concepts of family centred care and palliative care (Conner and Nelson 1999, Heyland, Rocker et al. 2002, Ahern 2013, Hellmann 2014). This is based on focusing care on the child–parent dyad (Fenwick et al., 2008), respectively on the 'family-professional partnership' (Kenner, Press et al. 2015, p. S20). Thus, in addition to communicating treatment options to parents this includes creating moments and spaces for parents and children to experience themselves as families, appreciating the child's life, making memories and, when the time comes, supporting parents to let their child go and acknowledge different manifestations of grief, and (Ahern 2013, Stafford 2015, Abraham, Battaglia et al. 2016). Any lack of transparency in information or continuity of care and/or insensitive verbal and non-verbal communication can challenge this and, as shown in our study as well, parents heavily depend on HCPs communication skills and their willingness to help and provide such information (Thiele and Mader 2016).

Critical Appraisal of the Studies

There are several strengths and limitations of the present thesis. I will shortly review the three successive studies and its main limitations. First, a general limitation of a telephone survey interview is that it does not allow for in-depth exploration and both the questions and responses are limited in their complexity. This also includes the complexity of the notion of solidarity. Due to the time constraints of a telephone survey, solidarity could not be clearly defined to respondents prior to the survey. Therefore, it is most likely that respondents may have understood only the most general notion of the term.

Second, the parental interview study aimed to shed light on the perspectives of parents regarding end-of-life decision-making. Although we aimed to include a large variation of cases throughout 2013–2015, the total number of 13 cases (i.e. 20 parents) from 2013-2014 was relatively low. Moreover, our study was limited to one University Hospital. Consequently, the experiences of the parents in our study are not necessarily representative of the experiences of other parents in the same situations and the small size and qualitative design limits the generalizability of the results.

Third, an analysis regarding threshold policies aimed to explore the ethical reasons and justifications. The study, however, may be biased by its North American and Northern European perspective. The literature on threshold policies is mostly influenced by the North American context (Janvier, Bellieni et al. 2016, Dupont-Thibodeau, Hindie et al. 2017), it is not clear how this translates to the whole of Europe (Hagen, Therkelsen et al. 2012, Hansen, Janvier et al. 2013, Janvier and Lantos 2016, Laventhal, Verhagen et al. 2016), and in particular to Switzerland (Berger, Steurer et al. 2017). Additionally, this ethical analysis led to the exploration of six rationales for such policies, which may not be an exhaustive list of arguments.

I see two main strengths of the present thesis. First, the structure of the studies in which the outcomes of the population survey enabled a further in-depth reflection on the actual experiences of parents. Second, the investigation of the perspectives of parents who lost an infant, as a vulnerable and less investigated population (Moro, Kavanaugh et al. 2006, Moro, Kavanaugh et al. 2011), is very valuable. In the next paragraph, I will illustrate that further research is needed to investigate or corroborate some of the mentioned limitations in the three study components.

Implications in Practice and Future Research

There is still much to be learned in end-of-life decision-making for extreme preterm infants about how parents and health care professionals make decisions in practice and on how these decisions should be conceived from an ethical perspective. This research projects leads to some potential ideas for future practice and research in neonatal ethics.

Share in Decisions to Continue or Discontinue Life-Sustaining Interventions

Parents should be given the opportunity to share in the decision whether to continue or discontinue treatment for their child. I recommend a shared approach, which extends the practice of asking parents for their agreement. Hence, parents should be continuously involved in re-evaluating the appropriateness of LSIs and it should be re-evaluated whether they want to share in the decision-making or not. Decisions must integrate the values of families and be taken together with a balance of the benefits and burden of the LSIs. This means allowing the broadest range of reasonable options without infringing upon the interests of parents or infants (Swinton and Lantos 2010, Batton and Batton 2013).

Since both prognosis and diagnosis are often ambiguous for this patient population (Wilkinson, Thiele et al. 2012, Marc-Aurele and English 2017), it is important for HCPs to openly communicate such prognostic insecurities. This is necessary to prepare parents for a spectrum of possible outcomes such as death during labour or at some postnatal stage, which requires parallel planning, i.e. “plan for survival and prepare for death” (Mancini 2017). Hence, transparent communication strategies – such as sharing weighted information, discussing a range of treatment options, providing parents with time to think and discuss, and building trusting relationships through the continuation of care – can enable parents to participate in decision-making based on their preferred level of involvement (Daboval and Shidler 2014, Hendriks and Abraham 2017). It should be respected when parents prefer not to (completely) participate in the decision-making and wish to be guided by the health care professional’s recommendation. Furthermore, it may be possible that some parents need more guidance and support in the beginning of the decision-making process. Either way, HCPs should continuously inform parents and aim to ensure they understand the available medical information. In addition, EoLDM processes should be precisely documented in the hospital charts and parents’ experiences should be systematically collected in order to get the feedback necessary for continuous improvement.

Further Institutionalize Palliative and Bereavement Care in the NICU

Our results illustrate the importance of further institutionalizing palliative care (PC) in neonatology – a holistic approach that starts at the moment of recognition of a possible palliative condition,[4] and provides supportive care during life and end-of-life for both the patient and the family. This enables "quality care" for the babies and "grief support" for the parents (Kenner et al., 2015, p. S19) and health care professionals (Garten and Hude 2014). The aim is not to prolong life at any cost, rather to ease the suffering of the patient, enhance his or her quality of life, facilitate informed decision-making with parents, and allow for a dying process in dignity and in such a way that is important for the family.

Establishing a good quality of PC is of particular importance for parents of extremely premature infants since parental bonding (i.e., physical, social, and emotional relationship-building interactions between parents and baby) has shown to be particularly difficult. Parental bonding might have to take place in parallel to anticipatory grief and to the impending loss of the child. Our study has identified this as an extreme burden for parents. The parents may only have a very short, but very precious time with their child since extremely premature infants may deteriorate and die more quickly than other patients (Williams, Munson et al. 2008, Einaudi, Le Coz et al. 2010, Tan, Docherty et al. 2012). Bereavement and perinatal PC further strives to minimize parental feelings of regret on how they spent the time with their child whilst alive and dying (Dickson 2017).

Perinatal PC has gained increased interest over the last decade in Switzerland. Although there is a growing body of existing (inter-)national knowledge about key elements of perinatal PC and specific guidelines for its provision (Cignacco, Stoffel et al. 2004, SAMW 2013, Catlin, Brandon et al. 2015), evidence shows the inconsistency in the application of palliative care principles in this group of patients (Quinn and Gephart 2016, Marc-Aurele and English 2017). Implementation of palliative care services remains especially low among neonates (Keele, Keenan et al. 2013). One study conducted in Switzerland illustrated that only 7% of parents of neonates received specialized palliative care services (Zimmermann, Bergstraesser et al. 2016). Additionally, there is little data on how HCPs and parents actually proceed in the practice of perinatal PC. There is, therefore, a need to identify best practice, outcome measures and an evidence base for practice (Nicholl 2013). For this purpose, the University Hospital under analysis has submitted a research proposal that

[4] PC is often discussed beforehand in situations where there is a probability that a condition is incurable.

aims to prospectively evaluate and improve perinatal PC in Switzerland by systematically and thoroughly examining perinatal PC services and consultations to determine its utilization and efficacy.

Investigate How to Manage Divergent Attitudes and Values

Almost 20 years after the first investigations on attitudes and values regarding EoL decisions for extreme preterm infants, studies often reach similar conclusions (Lee, Penner et al. 1991, Norup 1998, Cuttini, Rebagliato et al. 1999, Rebagliato, Cuttini et al. 2000, Støre Brinchmann, Førde et al. 2002, Peerzada, Richardson et al. 2004). Studies suggest, on the one hand, the need for further investigation into the influence of attitudes and values of stakeholders on decisions (Boss, Hutton et al. 2008). On the other hand, studies hold that there is a need for more education about long-term outcomes in neonatology to educate HCPs who, in turn, can provide parents with better quality evidence and communication (Boss, Hutton et al. 2008, Lam, Wong et al. 2009, Dupont-Thibodeau, Hindie et al. 2017). The hope is that training HCPs to address parental values during the decision-making process may improve communication and leads to better decisions (Streiner, Saigal et al. 2001, Boss, Hutton et al. 2008, Janvier, Lantos et al. 2008, Lam, Wong et al. 2009).

When weighing the burdens and benefits of withholding or withdrawing life-sustaining measures, it is not always possible to avoid subjective judgments about harm, risk, and long-term benefit. Hence, improving the communication skills of HCPs might solve misunderstandings of the relevant facts. But as noted in the introduction, disagreements can be the result of a different understanding of the relevant facts *or because of different value*s (Larcher, Craig et al. 2015). Although it is important to avoid potential misunderstandings, reasonable people might disagree on the basis of different values. Tools such as decision-aids may help to reduce conflict and establish a better understanding of different values between HCPs and parents. Indeed, decision-aids aim to improve parental knowledge on the infant's condition, involve parents in the decision, and make a values-choice agreement. In turn, these aspects may facilitate shared decision-making (Kakkilaya, Groome et al. 2011, Guillén, Suh et al. 2012, Muthusamy, Leuthner et al. 2012, Moore, Lemyre et al. 2017). However, Haward et al. (2017) have pointed out some limitations of decision-aids. Although decision-aids provide parents with a comprehensive overview of medical information, they predominantly focus on informational needs. Complex deliberations between multiple interests seems to be – as of yet – outside the scope of decision-aids (Haward, Gaucher et

al. 2017). In addition, the information presented in decisions-aids are based on population statistics, which does not refer to the individual outcome for a specific infant. This might be difficult for parents to understand and distinguish. The tool of communicative ethics from Jürgen Habermas, as adapted by Daboval et al. (2014) to the neonatal setting, aims to address the often overlooked personal experiences and value system of parents and HCPs, which may shape their interpretation of medical data and moral judgements on what is in the best interest of the infant. According to Daboval et al. communicative ethics proposes 'to recognize and promote all actors' participation in the discussion; to recognize the differences among actors; and importantly, to accept that everyone is morally equivalent to one another' (Daboval and Shidler 2014, p. 303). However, from a clinical perspective the question is whether one can always be morally equivalent and whether rare situations necessitate an exception.

Hence, future research should investigate the empirical value of communicative ethics and the possibility to alter existing decision-aids in order to include a recognition and deliberation of different experiences, attitudes and values between parents and HCPs. Openly acknowledging and discussing the attitudes, values and potential biases among stakeholders is an important step towards transparent ethical care of neonates and is pertinent to examining the moral validity of attitudes (Dupont-Thibodeau, Hindie et al. 2017).

Explore whether Transparent Counselling is Warranted in Withholding Treatment

As shown, transparent communication, empathy, and honesty play an important role in building a trusting relationship with HCPs (Brosig, Pierucci et al. 2007, Obeidat, Bond et al. 2009, Xafis, Wilkinson et al. 2015, Zimmermann, Bergstraesser et al. 2016). In knowing this, one might question whether HCPs should make explicit and transparent the national or hospitals' recommendations to extreme preterm infants at the limit of viability and disclose, before or after birth, the reasons behind such management decisions. Do HCPs have a duty to disclose local or national reasons toward resuscitation decisions of extreme preterm infants? Some might argue against informing parents about such policies, since this might unnecessarily burden parents. The knowledge their child was close to the threshold might not alleviate but rather cause harm. But, as previously shown, open and honest communication – albeit provided in a sensitive manner – can help parents to better cope with uncertainty, feel more in control, feel less anger and distress, which can lead to building trusting relationships with HCPs (Kavanaugh 1997, Sharman, Meert et al. 2005, Davies, Contro et al. 2010, Branchett and Stretton 2012, Ruthford, Ruthford et al. 2017).

Although it is possible that nondisclosure happens with the best intentions, i.e. keeping the well-being of parents in mind, unforeseen disclosure may cause harm and must be taken seriously (Dierickx 2007, Meadow and Lantos 2011). Future research should explore (non-)disclosure of resuscitation policies and whether 'speech is silver or silence is golden' from an ethical but also empirical perspective. But future research should also explore how much silence or how much information should be given to parents. Thus, it should be further explored how much information should be shared within a shared decision-making process and a further exploration of the duty to disclose regarding gestational age policies is warranted.

Investigate Practices at the Limit of Viability

Our parental study suggests that the infants born at 23 completed weeks were denied life-sustaining interventions. This raises the question whether infants born at 23 completed weeks were systematically denied LSIs in the University Hospital under analysis and whether this is the case in the whole of Switzerland. Nevertheless, even though we think that the richness and thickness of the qualitative data from the parental interview study outweighed the small sample size, our explanatory model was hypothetical in nature and needs to be critically assessed in further studies with prospective and observational designs (O'reilly and Parker 2013). Hence, future observational or ethnographic studies are necessary to explore these management decisions more closely at the bedside.

In addition, this should not only be explored in-depth in Switzerland, but is also warranted in other countries in North America and Europe. Although concerns regarding gestational age policies are voiced by parents and clinicians and investigated in many questionnaire studies (Janvier, Leblanc et al. 2006, Staub, Baardsnes et al. 2014, Dupont-Thibodeau, Hindie et al. 2017, Marmion 2017, Ruthford, Ruthford et al. 2017), little ethnographic data exists from clinical practice (Christoffersen-Deb 2012).

Normative Implications

There are also some normative implications based on the findings from the third study component of the present thesis. It is important to note, that these reflections and recommendations are not basing claims on the empirical data of the first two study components, the reflections are rather based on the ethical analyses of threshold policies.

Question Thresholds in Policies for Decisions on Resuscitation

The third study component of the present thesis has shown that policies that rely heavily on gestational age deny some babies treatment that might be beneficial. We have argued that policies that limit life-sustaining treatment based on the estimated gestation of an extremely premature baby are neither medically sound nor just. Based on the arguments provided in the third study, I support that parents of extremely premature infants should be appropriately informed to make an individualized and shared decision with health care professionals regarding life-sustaining interventions. Such a clinical decision should be based on an individualized estimation of the likelihood that treatment would be beneficial.

First, I hold the view that current methods of estimating gestational age are not sufficiently accurate for this to be the sole basis for decision-making (Batton and Batton 2013, Papageorghiou, Kemp et al. 2016). This is especially relevant in North America where it is common to assess gestational age with the history of the last menstrual period leaving a GA estimation with a broad range of -6 to +14 days (Wingate, Alexander et al. 2007, Butt, Lim et al. 2014, Skupski, Owen et al. 2017). But this is also relevant in most European countries, including Switzerland, where GA is estimated by early ultrasound in which the precision is ± 4 days (Sabbagha 1987, Berger, Bernet et al. 2011, Papageorghiou, Kemp et al.). This might seem of little value but when applied as the sole criterion it can affect a life or death decision. Even with the most precise measure to date, an early ultrasound measure, the gestational age of a pregnant women with an estimated GA of 23 weeks and 4 days (not eligible for care unless positive prognostic factors) could range from 22 weeks and 6 days (not eligible for care) to 24 weeks and 2 days (eligible for care). Yet we are dealing with the same mother, the same family and the same unborn infant.

Second, decisions, and thus policies, should be based on the most accurate, objective and individual prognostication that takes into account all relevant clinical characteristics. Then, prognostication should include both national as well as local statistics. Otherwise, policies could be based on a self-fulfilling prophecy. Even though there might be valid reasons for not treating infants born ≤24 weeks GA, there is a risk of a self-fulfilling prophecy when a national recommendation or hospital denies life-sustaining measures at, for example, 22 weeks because they never had a survivor. This reasoning is circular: '[w]e never try because it never works. And, it might be that it never works because we never try' (Mercurio 2005). For this reason, it is important

that parents are counselled about the local but also the (inter-)national chances of survival in making a resuscitation decision at the borderline of viability.

However, some ethicists have argued that such gestational age-based guidelines 'are the best available way of providing a structure for those involved in perinatal decision making, setting out the boundaries of appropriate care, and supporting parents and clinicians facing extremely difficult choices' (Wilkinson 2012). Without such structures, decisions would become subjective and risk inconsistencies (Wilkinson 2012, Marlow and Wyatt 2016). Likely so, but, as pointed out by other ethicists, consistency in these decisions is not by itself a worthwhile goal. It is possible to be morally wrong. For example, it is morally problematic that a mother presenting with the same high-risk pregnancy to two hospitals with the same capacity to intervene, yet different EGA-focused policies, will guide decisions in two opposed directions (Janvier, Barrington et al. 2012). This is problematic because decisions are based on an imprecise notion of gestational age, disregard other (potentially positive or negative) prognostic factors and/or are based on local outcome data. It is, therefore, important to find the right balance between 'consistency and local ethos' that should be based on sound argumentation (Janvier, Barrington et al. 2012). We have known from the beginning that in the realm of uncertainty reasonable people can disagree about reasonable measures. Although EGA-focused policies represent a comfortable option by offering an easy and straightforward method, simple rules are not adequate for these difficult decisions and cannot be ethically justified (Janvier, Barrington et al. 2008, Swinton and Lantos 2010, Janvier and Lantos 2011). Counselling expectant parents should start with a balanced risk assessment of the infants' estimated gestational age (uncertain), estimated birth weight (very uncertain), place of birth, usage of antenatal steroids, single or multiple birth, and should then explore the family's' values and wishes. Parents together with HCPs must decide on a course of treatment that reflects the values and desires of parents while incorporating the best interest of the child. This can potentially present challenges but establishing individualized goals of care with families of extreme preterm infants is morally superior from labelling infants in a fixed gestational age category (Dupont-Thibodeau, Barrington et al. 2014, Boland, Davis et al. 2016, Ruthford, Ruthford et al. 2017). Additionally, it has the advantage of being more flexible and transparent, while potentially reducing morally troublesome inconsistencies between centres and HCPs (Gillam, Wilkinson et al. 2017).

Although I defend the view that resuscitation decisions should be based on an individualized assessment of the infant and familial context, such changes cannot happen overnight. Changing

policies is but one step in a long process of cultural change. Consequently, it is necessary to continue to address and discuss the limits of gestational age estimations and local outcome data within the neonatal community to foster a change in the way physicians treat babies born extremely premature. It is furthermore important to realize that thresholds of care can change with time, technology, politics (i.e. national recommendations), economics and morality (Lantos and Meadow 2009). Therefore, obstetricians, neonatologists, midwifes, neonatal nurses and ethicists should continuously reflect and debate on the boundaries of the grey zone, realizing that these boundaries are not static.

Conclusions of the Thesis

In summary, the present thesis illustrated the epidemiological outcome data on survival and long-term development. Facts and values are interconnected and can be biased by research limitations (e.g. denominator bias) and/or attitudes and values. This illustrates the importance of being as explicit as possible in the communication with parents during end-of-life decision-making. On the one hand, explicitly take into consideration the research limitations affecting survival and long-term outcomes, and on the other hand, bear in mind that differences in attitudes and values can affect decisions at the end-of-life. I furthermore presented an overview of the ethical principles – best interest principle, futility, quality of life, parental authority, health care professional duties and shared decision-making – relevant to EoL decision-making for extreme preterm infants.

Based on the empirical findings of the present thesis, I propose and discuss several potential ideas for future practice and research in neonatal ethics. First, I recommend shared decision-making in decisions to continue or discontinue life-sustaining interventions. Parents should be given the opportunity to share in the decision whether to continue or discontinue treatment for their child through transparent and high quality communication strategies. Second, our results illustrate the importance of further institutionalizing palliative care (PC) in neonatology and further research best practice, outcome measures and an evidence base for practice. Third, future research should investigate how to manage divergent attitudes and values. Tools such as decision-aids or a communicative ethics may help to reduce conflict and establish a better understanding of different values between HCPs and parents. Fourth, the knowledge that open and honest communication provided can help parents to better cope with the situation necessitates further exploration of (non)disclosure of resuscitation policies. Fifth, future observational or ethnographic studies are necessary to explore management decisions at the limit of viability more closely at the bedside.

Based on the normative findings of the thesis, I urge that parents of extremely premature infants should be appropriately informed to make an individualized and shared decision with health care professionals regarding life-sustaining interventions, with clinical decisions based on an individualized estimation of likelihood that treatment would be beneficial.

In conclusion, although the beginning and the end of the lives of extremely premature babies are so close, we should support these infants and their families to make the decision that is right for the infant and his or her family.

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2003 – 2008	Secondary Education, Willem de Zwijger College, Bussum, Netherlands
2008 – 2012	Bachelor of Cultural Anthropology (BSc), University of Amsterdam, Netherlands
2012 – 2013	Master of Science in Medical Anthropology (MSc), University of Amsterdam, Netherlands
2013 – 2014	Master of Science in Bioethics (MSc), University of Leuven, Belgium
2014 – 2017	Doctoral Program in Biomedical Ethics (PhD), the Institute of Biomedical Ethics and History of Medicine, University of Zurich, Switzerland

EMPLOYMENT HISTORY

02/2012 – 06/2012	Research internship at the Right-to-Die Association in the Netherlands, Advisor: Dr. Anja Hiddinga, *Department of Anthropology and Sociology, University of Amsterdam*
01/2011 – 07/2013	Web-Editor 'This is the End', Employee: Marleen Peters, *Right-to-Die Netherlands*
2/2016 – 12/2016	Research Assistant for Validation of the Bernese Pain Scale for Neonates with Consideration for Contextual Factors (SNSF 159573), Advisor: Eva Cignacco, *Bern University of Applied Sciences*
09/2014 – 10/2017	Doctoral Researcher on factors in decision-making for extremely preterm infants (SNSF NFP67), Advisor: Dr. med. Jurg Streuli, *Institute of Biomedical Ethics and History of Medicine, University of Zurich*
05/2017 – Present	Research Assistant for Database of Individual Patient Experiences (DIPEx), *Institute of Biomedical Ethics and History of Medicine, University of Zurich*